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BACK PAIN DUE TO LOOSE FACETS OF THE LOWER LUMBAR VERTEBRAE

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Received for publication April 4, 1955

One of the causes of sudden severe low back pain is the unattached facet. This derangement of the back usually produces episodic attacks of excruciating discomfort, characterized by sudden onset and equally rapid relief of symptoms. However, this pain pattern is not unique to this disorder nor do the patients complain in a consistent manner; therefore, the history, physical and operative findings of six patients with this back derangement will be presented.

Case 1. A 47 year old white female complained of intermittent low backaches of two years duration. Over time this pain had become more acute. It radiated across the low back towards the lateral aspect of the right thigh. There was no history of trauma.

The patient stated clearly that she could flex her back without difficulty, but it was painful to come erect or to hyperextend it. When performing these motions, she often felt a "clicking" sensation in the low back. Generally rest would relieve the back pain, but not completely. Only when she felt "something move" would she feel better. Coughing and sneezing aggravated the pain when it was present. Hospital admission was necessary because of a very sharp attack of low back pain that had come on suddenly a few days before and had not been relieved by rest and the liberal use of narcotics.

On examination the patient was found to be in considerable distress. She very definitely resented having to make any kind of movement. The paravertebral lumbar muscles were in marked spasm and were very tender to palpation. The trunk had a tilt to the right. Flexion of the back was limited to a few degrees. Attempting to bend forward was not at all troublesome but when extension of the back was tried, the pain was excruciating. There was pronounced tenderness on percussion over the third and fourth lumbar spinous processes.

Except for the difficulty in moving the back, very little else in the way of physical findings was present. There were no reflex, motor or sensory changes in the legs. The straight leg raising test was positive on the right at 80 degrees, producing principally pain in the back.

X-rays of the back, including anterior-posterior, lateral and oblique views failed to demonstrate significant pathology. A myelogram was normal.

Because of the marked discomfort, it was decided to explore the lower lumbar spine. The intervertebral discs of the L_3, L_4 and L_5 interspaces were examined and found normal. Each facet was then inspected and to our surprise the inferior facet of L_4 was found separate from the lamina. It was not in its usual position, but had rotated medially and become wedged between the lamina and ligamentum flavum. It was easily removed, being attached to the soft tissues of the back by remnants of the ligamentous capsule. The free surfaces of bone were eburnated and a definite articulating surface could no longer be identified.

The postoperative course was uncomplicated. Most of the back pain was immediately

relieved and when the patient was discharged one week later there were very few complaints. It has now been a year since operation during which time the patient has been actively running her household without the appearance of any untoward symptoms.

Comment: Review of the x-ray films after operation still failed to demonstrate the lesion found at surgery. Pathologic study of the removed fragment of bone was not helpful. Whether the bone had been a congenital malunion of the facet or whether it had been fractured years before, no one could tell. The only thing that seemed clear was that the patient's back contained a facet not attached to its proper neural arch which at certain times interfered with its proper motion, impinging on sensitive structures and resulting in marked discomfort.

The question of etiology of many free facets cannot be settled fully because there are evidences for both developmental defects and the result of trauma. The case history outlined below seems more clearly an example of the former.

FIG. 1. Case 2. Note linear defect in neural arch of L_4.

FIG. 2. Case 2. Anterior-posterior projection fails to demonstrate any definite lesion in this area.

Case 2. A white male, 31 years old and a day laborer was admitted to the Robert Packer Hospital for evaluation of his back complaints. For nine years he had experienced low backaches which were most persistent. On certain, but unspecified movements, he said "something moves" or "gives out" in his back. This was followed by an immediate and exceedingly severe back pain so that he was unable to move. The attacks lasted for several days, then subsided quickly and he was able to return to heavy work immediately thereafter. He had found that walking, riding machinery, handling logs, lifting, etc., might initiate an attack. He has been having several such episodes yearly. On rare occasions pain had radiated into the sacral region and down the backs of the legs to the ankles.

Physical examination during the attacks was reported as showing tremendous spasm of the paravertebral lumbar muscles and inability to move the back in any direction without marked pain. Several examiners had expressed surprise when the patient's symptoms suddenly evaporated a few days later. The diagnostic impressions on previous hospital admissions include the terms "hysterical" and/or a "malingerer."

When seen during a quiescent period the only physical finding was tenderness on pressure over the L_3 and L_4 spinous processes. X-rays of the back, on the lateral projection, showed

FIG. 3. Case 2. Oblique view shows defect near inferior facet of L_3. The pars interarticularis is intact.

a linear defect of the neural arch of L_3 but not through the pars interarticularis or pedicles. Just what this line meant was not correctly interpreted by the many physicians who saw the films. Myelography was not helpful.

At operation, both inferior facets of L_3 were found attached to the lamina by dense fibrous tissue only. False joints were present. The facets were easily removed and were found to overlie the L_4 nerve roots. The spinous process, lamina, pars interarticularis, superior facets and pedicles of the neural arch were intact. There was no abnormal mobility of the body of the L_3 vertebra. A small amount of bone was rongeured off the lamina to prevent it striking the L_4 vertebrae on extension.

The postoperative course was uneventful, the patient leaving the hospital on the seventh postoperative day. He returned to heavy work about six weeks later. He has had no further trouble with his back.

Comment: The significance of the defect on the x-ray films had been overlooked by previous examiners because it appeared in only one projection. The accompanying illustration attempts to demonstrate it. Because the loose

FIG. 4. Case 2. Postoperative film shows clearly the bilateral symmetrical removal of the interior facets of L_3.

facets were so symmetrical and because of traces of cartilaginous-like material in the false joint, the lesion appears to be a developmental fault.

Trauma, on the other hand, can definitely produce an isolated fracture between the lamina and inferior facet. This apparently need not be severe. If a large defect in the lamina, near the facet, is made during the operation for removal of an extruded intervertebral disc, the facet may break loose postoperatively with only mild trauma such as an unguarded motion of the spine.

Case 3. A 24 year old white woman was admitted to the hospital because of recurrence of severe back pain and discomfort in both legs. She has always been obese and while her height is only five feet, three inches, she weighs 250 pounds. In 1950 she had been operated upon for a ruptured intervertebral disc at L_4. The postoperative course had been smooth and she had remained well until February 1952 when she slipped on a patch of oil and twisted her back. Following this she complained of low backache and sciatica. She was seen as an outpatient on several occasions when it was noted that motion of the back was lim-

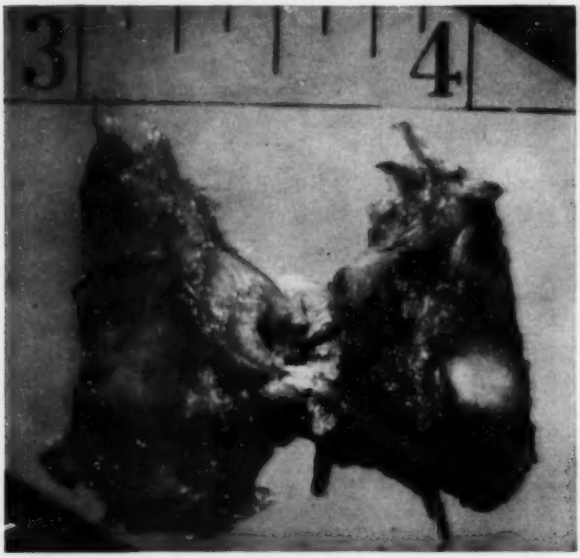

FIG. 5. Case 2. Facets removed at operation, viewed from same projection as seen by the operator.

ited and that she was moderately uncomfortable. Conservative measures were tried and weight reduction attempted, both with conspicuous lack of success.

During May, 1952 she fell down some stairs, again wrenching her back and striking her head hard enough to produce unconsciousness. The back and leg pain increased considerably following this accident. On admission to the hospital, the patient was very uncomfortable with low back pain even during bedrest. There was pronounced tenderness on pressure of the lower lumbar spinous processes. The straight leg raising test was positive at 45 degrees on the right and 30 degrees on the left. The right ankle jerk was absent and the left depressed. Hypalgesia was noted over the lateral aspects of both calves. X-rays of the lumbar spine showed only a narrowing of the L_4 and L_5 interspaces.

Bed rest in the semi-Fowler's position and Williams exercises were instituted. There was very little reduction of pain and after a week of this therapy she was allowed to sit up briefly which immediately resulted in an acute exacerbation of symptoms. At operation, it was found that the left inferior facet of L_4 had been fractured and was lying on the L_5 root. The facet was removed and an intervertebral fusion done. The postoperative course was uneventful and the patient was discharged at the end of ten days. The patient has been seen at frequent intervals. There has been no reduction in obesity. She complains of low backaches but remains active enough to run a household and drive a taxi. There have been no recurrences of acute back pain.

Comment: Because of the obesity, a longer-than-usual exposure of the interlamina space was made at the first operation. So much of the lamina near the inferior facet had been rongeured away that the facet snapped off and became a loose body following the trauma of falling. From the history and the marked

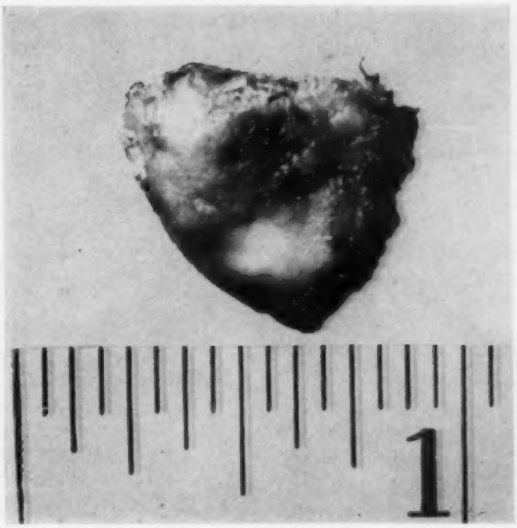

FIG. 6. Case 4. Free inferior facet at L_4.

increase of back pain following the second fall, it seems likely that the fracture was sustained at that time.

We have recently encountered another instance where this has happened following operation for a protruding intervertebral disc. Here the exact time of fracture is not easily determined.

Case 4. This patient is an 18 year old girl who entered the hospital in November, 1953 with the typical signs and symptoms of ruptured intervertebral disc. She was five feet, two inches tall and weighed 165 pounds. Conservative measures used to relieve the symptoms failed. Operation was undertaken, but due to the obesity and anatomical position of the intervertebral disc, more than half of the lamina of L_4 was removed in order to inspect the L_4 disc. The protrusion was not at this interspace, however, and a herniated disc at L_5 was removed.

The immediate postoperative course was satisfactory but after the third postoperative day the girl once more complained of considerable backache and right sciatica. Throughout the next year periods of bed rest, back supports and corrective exercises were tried with no appreciable benefit. Her weight, however, was successfully reduced to 120 pounds.

Until November, 1954 pronounced tenderness of the back persisted being most marked just to the right of L_4 spinous process. Flexion of the back was limited to 45 degrees and hyperextension was very painful. There was a definite limp of the right leg on walking and dorsiflexion of this foot was poorly performed indicating considerable weakness of the anterior tibial and peroneal muscles. There was an extensive area of hypesthesia involving the lateral and posterior calf and almost the entire foot on the right, indicating damage to the roots supplying the L_5 and S_1 dermatomes. X-rays of the back showed a normal lumbar curve and no evidence of fractures of the lamina.

At operation, the inferior facet of L_4 was found broken at the lamina and lying on the

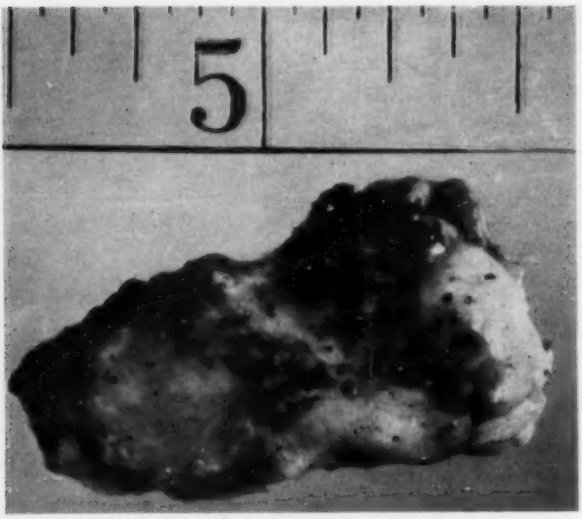

FIG. 7. Case 5. Free inferior facet of L_5. Note eburnation of almost entire surface of loose piece of bone.

L_5 root. The facet was easily removed and the opposing surfaces at the fracture site were eburnated. A soft protruding disc was also present and this likewise was removed. The postoperative course was very smooth and the back and leg pain have subsided. Some strength has returned to the dorsiflexors of the foot but the sensory changes persist.

Comment: Again the attachment of the facet to the lamina had been so weakened by removal of bone at the first operation that the facet became detached after only mild trauma. Its presence directly over the L_5 root, coupled with a protruding disc at this interspace can easily account for the failure of symptoms to subside. Nerve roots can often accommodate themselves to protruding discs but the intermittent trauma produced by a free bony facet is not so easily tolerated.

Whether a facet can be traumatically fractured from an intact lamina as an isolated break cannot be stated with certainty. In the patient whose case history is related below, it would appear that the facet had been loose for a great number of years, and may possibly have been the result of trauma.

Case 5. This 48 year old man injured his back in an automobile accident thirty years before, requiring hospitalization for 14 weeks. Information concerning the exact nature of the injury is not available. Ever since that time he has been subject to recurrent backaches and has been careful when lifting and handling heavy objects. There had never been any discomfort in the legs.

A few days before the patient was first seen by us on May 20th, 1953 he had developed a much more severe back pain than usual and the pain quickly radiated across the left

buttock and down the posterior thigh and to the knee. The pain soon became excruciating and he was unable to lie down or move in any direction. The only relief that could be obtained was by sitting. Liberal amounts of morphine and Demerol afforded no relief. He sat so still for several days that his legs had become swollen and red.

On examination, the patient was in marked distress with constant stabbing back pain. On attempting to stand, there was a pronounced tilt to the right. He stood flexed forward about 30 degrees and he resented having to place any weight on his feet. The paravertebral lumbar muscles were in marked spasm. Back pain was unbearable when attempts to try the straight leg raising test were made. Pressure on the L_3 and L_4 spinous processes aggravated the back and leg pain. There were no reflex or sensory changes in the legs. X-rays of the lumbo-sacral spine showed osteoarthritic changes about the L_3 and L_4 vertebrae, indicating trauma in the past about this area. Our clinical impression was that the patient had extruded an intervertebral disc.

At operation, the discs at L_3, L_4 and L_5 were not protruding. The facet at L_3 of the left was loose, had rotated under the lamina and lay over the L_3 root. It was surrounded by dense fibrous tissue but was easily removed. Definite articulating surfaces could not be distinguished and the bony fragment gave the appearance of having been detached for a long period of time.

The postoperative course was very smooth. All the back pain was gone by the fifth postoperative day. When discharged on the tenth postoperative day, he was walking without difficulty, the back was straight and he could flex it 30 degrees without discomfort. He has returned to running his farm and has had no back pain for the past 18 months.

Comment: Whether this patient had a unilateral anomaly similar to the patient discussed in Case 2, which became dislocated during the automobile accident, or whether the facet was actually broken off an intact lamina cannot be stated categorically.

When the bony structures were seen 30 years after the trauma, the lamina looked normal and we could tell nothing from the loose piece of bone removed as the normal landmarks of a facet had been obliterated by a combination of rubbing and absorption. We can only say that it must have been the seat of his troubles as this past year has been the first in thirty that his back has not bothered him.

In the final case discussed, trauma appears to be the initiating cause but again a developmental peculiarity cannot be eliminated.

Case 6. A well developed 35 year old man was first seen by us on October 13, 1952. He related that while he was pulling on a wrench on May 13, 1950, it had slipped and he fell backward "snapping" his back. An immediate, rather severe low backache was experienced which "laid him up" for two weeks. Since then he had been unable to do heavy work, and was subject to intermittent low back pain, radiating toward the right anterior thigh. Walking, sitting and bending were particularly bothersome. He experienced "catches" in his back, especially on getting up in the morning and he had considerable difficulty in getting out of bed. There had been several episodes of numbness and tingling in the anterior aspect of the right thigh. Coughing and sneezing often aggravated the back pain. On a few occasions there had been pain down the right leg as far as the lateral aspect of the calf.

Examination disclosed that all motion of the back was above L_4. The patient could flex

well without difficulty, but it was painful to come erect. The lordosis in the low lumbar region persisted when the patient flexed his trunk, but motion remained in the upper lumbar vertebra. The paravertebral muscles became very tense and the patient gave a "jerk" to get himself erect. Hyperextension and rotation were limited and painful. Pressure applied over the L_4 and L_5 spinous processes was very uncomfortable. There was no weakness of the legs, no reflex or sensory changes. Straight leg raising caused marked back pain bilaterally at 30 degrees. Detailed x-ray examination of the back, including a myelogram was normal. Our clinical impression was an "unstable" back (reason for instability not apparent) and a fusion was contemplated.

At operation it was found that the ligaments about the right inferior facet of L_4 had been avulsed. The cartilaginous surface of the L_4 facet together with a small portion of bone was separated from the lamina. This could easily be lifted out. The rest of the facet was rongeured off. There was no protrusion of the lower three intervertebral discs.

The postoperative course was smooth. The patient returned to work two months after operation. When last seen, two years after his discharge, he complained of mild backaches and occasional severe "cramps" in the right calf. He was working 40 hours a week in a factory and as a gasoline station attendant in the evenings and weekends. Rest would easily relieve the back discomfort. He could flex his back about 80 degrees but still came erect with a lumbar lordosis. Extension of the back was painful in the region of the L_4 spinous process. He feels very much better than he had before operation.

Comment: In this instance, the break had occurred at a somewhat different position than in the preceding cases. Here it was similar to an epiphyseal break, whereas in the former cases the break was several millimeters from the joint surface.

DISCUSSION

Very little appears in the standard orthopedic texts regarding the problem of loose facets. The general medical literature is also quite barren.

Two articles on this subject appeared in the same issue of the "Journal of Bone and Joint Surgery" for July, 1933. The first was by Nichols and Shiflett (1) who described the x-ray findings on seven patients on whom routine studies for backaches disclosed lesions identical with those under discussion. Six were men and one was a woman, with ages ranging from 25 to 57 years. Only two gave a history of trauma. Since the clinical symptoms and roentgenographic findings could not be correlated, the authors felt that the lesions could be explained only on the basis of a congenital malformation, namely, an ununited epiphysis or an atypical ossification.

The second article, by Mitchell (2), likewise describes the loose articular facet. In all of his patients there was a history of trauma and multiple loose facets were noted in three of the seven cases cited. "The fracture" was considered to be due to indirect violence following severe forward or lateral flexion associated with rotation. The presence of nonunion and the failure of callus formation was clearly recognized, being "explained" on the basis of lack of immobilization. Conservative measures resulted in symptomatic improve-

ment in some patients but long term follow-up studies were not mentioned. Operation with removal of the loose body followed by fusion was carried out in two patients whose symptoms had failed to subside.

In neither paper is there sufficient clinical, operative, histologic or x-ray evidence to in any way help resolve the dilemma as to whether we are dealing with ununited fractures of normal bones or congenital anomalies that have suddenly become symptomatic, or a combination of both. During the intervening 22 years, no clear illumination has been shed on this point.

Primary traumatic fractures must certainly be very rare. However, they may often be overlooked, since even when known to be present post-operatively it is often impossible to demonstrate them on preoperative x-rays. The occurrence of congenital anomalies of the neural arch with failure of fusion of the facets is better understood, although the incidence of this disorder is not precisely known either.

Present day practice leans so heavily on the x-ray in the diagnosis of bony disorders that many practitioners seem to have forgotten that not all pathologic processes are visualized by this means, even after the most detailed and exhaustive efforts. Our own experience obliges us to admit that inferior facets of L_3, L_4 and L_5 can be separated from the appropriate lamina and even be displaced from their normal articulating surface, without demonstrable x-ray evidence. The clinical picture of the patients described above clearly points to a mechanical problem in a specific area of the low back. Because the roentgen studies of the back were "normal", vague and poorly understood entities as "strains" and "sprains" were utilized as diagnoses. Even hysteria and malingering were considered suitable explanations for some of the symptoms. Only the conviction that some definite but unknown mechanical defect existed followed by exploration of the low back by surgery disclosed the cause of the symptoms.

From the review of the cases related, it will be seen that there is no definite symptom complex that covers all of them. Generally the back pain far exceeds the sciatic radiation if the latter be present. Patients may feel "clicks" or "something moving" in their backs. This is highly suggestive. Symptoms often come and go rapidly, especially with bending and extending the back.

Except for the pronounced muscle spasm during the episodes of acute pain, physical findings are few and unimpressive. When the roots of the sciatic nerve are impinged upon, the usual neurologic findings in the legs may be present. These, of course, are specific for location only, not for etiology. The history of sudden severe backaches that are intermittent is the most constant feature and should be an alerting signal for this condition. While the symptom complex displayed by these patients is not entirely unique, there are certain features that appear to separate it from the other common back disorders such as a herniated intervertebral disc, a free neural arch or spinal cord tumor. The abruptness of onset and recession of excruciating back pain is unusual

with any of the above conditions. None of the patients under discussion protested partially flexing the back in contradistinction to patients with a disc or free arch, both of whom find this maneuver difficult and painful. Hyperextension and coming erect produced an exacerbation of symptoms but by using the hands to help the movement, and by twisting themselves, patients were usually able to accomplish it. With other types of back disorders both flexion and extension are usually restricted and there are limits beyond which the patient cannot force himself. The severity of back pain experienced by these patients, accompanied by vague leg discomfort or mild sciatica, is unusual for free arches and protruding discs, but is common in cauda equina tumors.

It is thought that more of these chronically disabled patients with low back complaints should be thoroughly, widely and carefully explored when the signs and symptoms point to a mechanical disturbance, normal x-ray studies notwithstanding. The mechanical defect should be diligently sought at operation before definite treatment is decided upon. We are not yet at a stage where we can always make an adequate preoperative diagnosis and so what is to be done cannot be preconceived. Merely diagnosing an "unstable back" and covering up the pathology with a posterior fusion has hindered our understanding of the low back problem for years. We must admit to ourselves and to patients that medicine is not yet ready to explain all back pain despite our present armamentarium of logic, history, physical examination, laboratory studies, x-ray and intuition. I have yet to have a patient with severe backache refuse to let me explore his back when the problem was openly discussed and the risks not exaggerated even though nothing definite was promised.

The etiology for the production of the loose facets cannot be settled by the author. After reviewing several texts and articles both by anatomists and orthopedists, nothing but conflicting theories emerge, with evidence for and against all of them. The experience gleaned from operation would make me favor either a developmental anomaly or perhaps trauma very early in life. Except those where the lamina had been weakened at a previous operation, all the specimens removed were distorted, surrounded by heavy fibrous tissue and showed no evidence of even abortive fracture healing. It must be admitted, however, that those following a previous disc operation also failed to show healing though the articular portion of the facet appeared normal. This inability to be certain of the etiology makes the problem of compensation difficult where liability exists. The author has been unable to answer this question either to himself or to inquiring lawyers.

Very little by way of precedent can be established for the proper form of treatment. Prolonged immobilization or support has been advocated with a fusion in late (whatever these may be) cases or those in which conservative measures fail. Neither of these methods would appear to be entirely satisfac-

tory. It seems doubtful if the true fractures ever heal, while the anomalies (which are not always distinguishable by x-ray) certainly will not. The clinical symptoms may subside, but they will probably recur after varying intervals.

The most simple procedure seems to be to excise the facet. The operation results in a negligible morbidity and to date no mortality. Convalescence is rapid. If a facet has been loose for years, it has contributed nothing to the stability of the back. Fusion can be done later, if symptoms warrant. To date we have had no reason to resort to this procedure.

SUMMARY

Six cases in which inferior facets were separated from their adjacent lamina are related. Developmental anomalies probably account for most of them. The role of trauma is discussed. Some occur after the lamina has been weakened by previous surgery. The difficulty at arriving at a correct diagnosis preoperatively is discussed, especially the failure of x-rays consistently to demonstrate the lesion. Simple excision of the loose facet appears adequate. A plea is entered for a more thorough surgical exploration of patients with marked local low back complaints, despite "negative" x-rays.

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HERMAPHRODITISM: RECOMMENDATIONS CONCERNING ASSIGNMENT OF SEX, CHANGE OF SEX, AND PSYCHOLOGIC MANAGEMENT*

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CRITERIA OF SEX

The homespun wisdom of medically unsophisticated people confronted with a newborn hermaphrodite usually guides them to assign the baby to the sex which it most resembles in external genital appearance. This procedure is, after all, only an extension of the age old practice of inferring, on the basis of a single glance, that the reproductive system in its entirety is either masculine or feminine. Before the advent of modern surgical and miscroscopic techniques, there was no way better than the homespun one of deciding the sex to assign to an hermaphrodite. After the advent of modern surgery and microscopy, it was assumed that gonadal structure as revealed by microscopic examination of sections of gonadal tissue, was the ultimate criterion to use in assigning the sex of an hermaphrodite.

This micrsocopic criterion was the basis of Klebs' well known classification of hermaphrodites in 1876 (1). Klebs recognized true hermaphrodites who possessed both ovarian and testicular tissue, male pseudohermaphrodites with only testicular, and female pseudohermaphrodites with only ovarian tissue. It was widely assumed that gonadal structure would determine sexual outlook and desires, even in those cases where gonadal structure and secondary sexual characteristics at puberty were paradoxically contradictory. Thus, it was taken for granted that the microscope would reveal the real sex of pseudohermaphrodites and decide the issue of their rearing. Only when the microscope revealed true hermaphroditism did the physician have an open choice in assigning the sex of rearing.

Klebs' archaic classification has survived despite the fact that it was made before anything was known of endocrine secretions and sex hormones; before any extensive psychologic research had been done on the determinants of the psychologic phenomena of sexual outlook and orientation; and before a reliable

* These recommendations are based on a study of psychologic development in 65 hermaphroditic infants, children and adults, carried out during the past 3½ years at The Johns Hopkins Hospital. The study is supported by a grant from The Josiah Macy, Jr. Foundation, and is under the aegis of John C. Whitehorn, Professor of Psychiatry, and of Lawson Wilkins, Associate Professor of Pediatrics, Acting Pediatrician-in-Chief and director of pediatric endocrinology.

technique of chromosomal sex determination had been developed. Notwithstanding endocrine, psychologic and chromosomal discoveries, in the present day there are still many people, medical and lay, who unquestioningly accept the gonadal criterion, though the newly discovered technique of chromosomal sex determination from a skin biopsy (2, 3) has tempted some enthusiasts to adopt a chromosomal criterion instead.

Our studies of 65 ambiguously sexed people have demonstrated that it is extremely unwise to use a single criterion like gonadal structure or chromosomal pattern in assigning an hermaphrodite to one sex or the other. In view of what is currently known about the chromosomes of hermaphrodites and the contradictions which may exist between them and other signs of sex, the chromosomal criterion should be given a minor place and should never be used as the ultimate criterion.* Likewise with the gonads, the contradictions that may exist between them and other signs of sex are so great that the gonadal criterion should not be the exclusive one. The criteria to be more seriously considered and appraised for their relative importance are: external genital morphology, hormonal sex (with due recognition of the frequent difficulty of predicting hormonal sex before puberty and of the possibility of corrective hormonal intervention); and the gender role established and ingrained through years of living in a sex already assigned (5, 6).

By the term, gender role, we mean all those things that a person says or does to disclose himself or herself as having the status of boy or man, girl or woman, respectively. It includes, but is not restricted to sexuality in the sense of eroticism. A gender role is not established at birth, but is built up cumulatively through experiences encountered and transacted—through casual and unplanned learning, through explicit instruction and inculcation, and through spontaneous putting two and two together to make sometimes four and sometimes, erroneously, five.† In brief, a gender role is established in much the same way as is a native language.

From our studies of the life adjustments of the patients in our series, we have found it definitely advantageous for a child to have been reared so that a gender role was clearly defined and consistently maintained from the beginning. When, in deference to the presumed importance of gonads, a change of assigned

* Use of the chromosomal criterion alone would lead to absurdities in the case of, for example, patients formerly diagnosed as having ovarian agenesis. During the past year, it has been discovered that a large proportion of them are chromosomally male (4). They have entirely feminine external genitals and internal accessory organs. It is inconceivable that any physician would recommend such a person to live as a male.

† These statements do not represent adherence to a theory of environmental and social determinism, for experiences encountered do not dictate experiences transacted in a simple, point-for-point correlation. Transactions are frequently highly unpredictable, individualistic and eccentric, for reasons as yet not fully ascertained.

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sex was imposed later than early infancy, the life adjustment was not significantly improved and was often made worse.

VARIETIES OF HERMAPHRODITISM

In this paper, it is not our intention to expound at length on the differential diagnosis of the various types of hermaphroditism (7, 8) but it will be pertinent to list, with comments, the seven categories which were found to be both exhaustive and mutually exclusive in making a survey of over 300 hermaphrodites reported in the English language literature between 1895 and 1951 (9).

(*1*) *Female Pseudohermaphrodites with Hyperadrenocorticism*

The external genitals may appear almost normally female with slight to medium enlargement of the clitoris, or there may be a single urogenital orifice with enlargement of the clitoris; or, rarely, there may be a penile urethra and fused, empty scrotal sac. Physical growth and development is precocious and virilizing, but may be totally corrected with cortisone therapy (10). Very early postnatal diagnosis is possible by means of urinary 17-ketosteroid assessment, without exploratory laparotomy. Plural incidence in a family is common, but is usually restricted to a single generation and a single marriage. Gonads and chromosomal pattern are female.

(*2*) *Female Pseudohermaphrodites with Phallus, Normal Ovaries and Normal Mullerian Structures*

There may be a single urogenital orifice with penis-like clitoris and a single urethral meatus in the urogenital sinus; or the clitoris may be so penis-like as to be supplied with a urethra serving as an auxilliary urinary outlet. Pubertal development is entirely feminine and reproduction is possible. Once the chromosomes have been established as female, this condition can be differentiated from the former on the basis of urinary 17-ketosteroid measurements, but it cannot be differentiated on the basis of these two criteria alone from true hermaphroditism which, so far as is known, is the only other variety of hermaphroditism in which female chromosomes may be present. A complete diagnosis, therefore, requires laparotomy and gonadal biopsy. The condition is extremely rare.

(*3*) *True Hermaphrodites*

Ovarian and testicular tissue is present, either in one ovotestis, the other gonad being an ovary or a testis; in two ovotestes; or in one ovary and one testis. The nature of the chromosomes for all subvarieties has not been ascertained. In one case of true lateral hermaphroditism (left ovary, right descended testis, no gametogenesis) chromosomes were bilaterally female. Internal genital structures and external genitalia exhibit divers degrees of ambiguity. Pubertal development may be masculine, feminine or ambiguous. Diagnosis requires

exploratory laparotomy and biopsy of the gonds. No more than 60 cases have been reported in the European and American literature of the last century.

(4) Male Pseudohermaphrodites with Well Differentiated Mullerian Organs

The penis may be fully formed with penile urethra, or it may be hypospadiac. One or both testes may be cryptorchid. One testis may be atrophic. Mullerian structures often herniate into the groin or scrotum in the company of one testicle. Pubertal development is almost always masculine, sometimes rather eunuchoid, especially when cryptorchidism is bilateral and the penis hypospadiac. Diagnosis requires surgical exploration. Whether a female, as well as the usual male chromosomal pattern is compatible with this variety of hermaphroditism has yet to be ascertained.

(5) Simulant Females with Feminizing Testes

External genital appearance completely simulates the normal female. The vagina is a blind pouch and, with very rare exceptions, the mullerian system is a cord-like vestige. The wolffian system is malformed or vestigial. The testes remain in an ovarian position or herniate into the groin. Pubertal feminizing is complete, except for menses, absence of which is the first sign of this concealed form of hermaphroditism in otherwise apparently normal women, unless the testes have previously herniated. In some cases there is absence of pubic and axillary hair. Diagnosis requires biopsy of the testes which should not be removed since they produce estrogens and these patients always live as women. Familial incidence in several generations is common. The chromosomal pattern is male.

(6) Cryptorchid Hypospadiac Males with Feminizing Testes

These cases resemble those in the former group except that the phallus is slightly or greatly larger than a clitoris, and that the blind vaginal pouch, usually present, opens into the sinus of a single urogenital orifice. The mullerian system is vestigial, and the wolffian system malformed or vestigial. Pubertal development is feminine, but not always strongly so. It is impossible to predict this feminization beforehand, except in cases of familial incidence. In infancy and childhood, therefore, these cases cannot be distinguished from those of the next group. Diagnosis requires testicular biopsy. Plural incidence within a family may occur. The chromosomal pattern is male.

(7) Cryptorchid Hypospadiac Males

Historically, hypospadiacs have not been called hermaphrodites unless hypospadias is extreme, the scrotum bifid and the testes undescended. Careful scrutiny often reveals that such hypospadiacs have a blind vaginal pouch hidden beyond the single external urogenital orifice. There may also be a separate

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external orifice for a blind vaginal pouch. The phallus may be only slightly larger than a clitoris. A virilizing puberty cannot be predicted beforehand. Diagnosis requires testicular biopsy. Plural incidence within a family may occur. The chromosomal pattern is male.

SEX ASSIGNMENT: NEONATAL PERIOD

It should be the aim of the obstetrician and pediatrician to settle the sex of an hermaphroditic baby, once and for all, within the first few weeks of life, before establishment of a gender role gets far advanced. Some hermaphrodites are born with external genitals that look so completely masculine or feminine that ambisexuality is not suspected. The baby is unhesitatingly declared male or female and no question is raised perhaps for years. Early uncertainty is aroused only when the external genitals look ambiguous. It is our recommendation that, in assigning the sex in these ambiguous instances, consideration be given first to the appearance and morphology of the external genitals. If the external organs are so predominantly male, or so predominantly female that no amount of surgical reconstruction will convert them to serviceably and erotically sensitive organs of the other sex, then the sex of assignment should be dictated by the external genitals alone. All further surgical and hormonal endeavor should be directed toward maintaining the person in that sex.

If the external genital anatomy of a neonate is thoroughly ambiguous and the possibilities of surgical reconstruction are equally promising in either direction, then gonadal and hormonal considerations may be more heavily weighted with regard to sex of assignment. On the basis of gonadal structure alone, however, it is frequently impossible to predict a virilizing or feminizing puberty. In the particular instance of hyperadrenocrotical virilism in girls, it is possible to correct the hormonal incongruity by treatment with cortisone.

The chief objection to be raised against the recommendation of paying greatest attention to external genital anatomy in assigning the sex of a neonatal hermaphrodite is that the recommendation flagrantly disregards the issue of fertility. But this objection is more imaginary than real. No male pseudohermaphrodites or true hermaphrodites have been known to have become parents, and many have been proved sterile. Female pseudohermaphrodites, including hyperadrenocortical females regulated on cortisone, may become pregnant. When such female cases are diagnosed promptly at birth, it is precisely because of their obviously ambiguous genital appearance. Subsequent surgical feminization and hormonal regulation can be thoroughly successful, thereby preserving areas of erotic sensation, fortifying the feminine role and safeguarding the opportunity for eventual child bearing. The only exception to this generalization is the extremely rare case of a female pseudohermaphro-

dite with a fully developed penis and normal looking, though empty scrotum; but only an exceptional combination of circumstances would permit the diagnosis of female pseudohermaphroditism in such a baby in the neonatal period.

POSTNEONATAL PERIOD: CHANGE OF SEX

It is not in neonates, but in older infants and children that the most perplexing and difficult problems of sex assignment are encountered—problems of whether to change the sex already assigned. Even when fully aware of a baby's genital ambiguity, parents do assign it to one sex or the other before long. The language dictates that they refer to the child as he or she, and they cannot sit on the fence indefinitely before announcing the birth of a son or daughter. Using all the medical help available, parents make the best decision they can and thenceforth drift into the conviction that they have a son, or a daughter. They rear the child accordingly, despite the knowledge that they may have to make a change later, and the subtle processes by which a gender role is established have begun. The longer a change is postponed, the more difficult it becomes for parents to relinquish a son in favor of a daughter, or vice versa. After a change, there is a time lag, maybe an extensive one, as parents readapt themselves, which is not without effect on the child. Neighbors and friends also find it difficult to accept the change, and even more difficult to forget it. Later in life the child is likely to be confronted with coarse jokes and reminders, unless the family had started life entirely afresh in a new community at the time of the change.

Difficult though it be for parents and others to negotiate a child's change of sex, it is even more difficult for the child, once he or she has established a conviction of gender. It is not possible to state a fixed age at which gender awareness becomes established: as in other matters pertaining to development and maturation, it is not the same age for all infants. As a general guide, it may be said that the crucial age is somewhere around eighteen months. This claim is based on a study of 11 cases of change of sex; 4 of them were changed before nine months of age and subsequently assimilated their new gender role without identifiable signs of psychologic maladaptiveness. The other 7 were changed at or after fifteen months, the latest at sixteen years, voluntarily, and the other 6 before school age. All of these 7 had assimilated their new gender role in varying degrees of pervasiveness, but all except one, who was changed at two years and three months, subsequently evidenced at least one indisputable, chronic symptom of psychopathology. Severity of the symptom varied, though in no instance reached psychotic proportions. The incidence of psychologic maladaptiveness was conspicuously less frequent among cases with no history of change of sex.

Gender role is so well established in most children by the age of two and one-

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half years that it is then too late to make a change of sex with impunity. One must calculate the risk of ensuing psychologic disturbance. This risk may very occasionally seem worth taking, especially in cases of complete incongruity between assigned sex and external genital equipment, but psychiatric supervision and follow-up should never be omitted.

After the transition from infancy to childhood—which is another of those unfixed points in the developmental sequence, occurring some time between the ages of three and one-half years and four and one-half years—it is too late to impose a change of sex. In the exceptional instance of an hermaphroditic child who has privately construed that an error of sex assignment has been made, and who has secretly half-resolved on a change of sex, successful negotiation of a change may prove possible. But our experience has led us to believe that voluntary requests for change of sex in hermaphrodites belong to the teenage. Though such requests are rare, they deserve serious evaluation, for they are usually a culminating attempt to resolve years of well founded perplexity and doubt.

For neonatal and very young infant hermaphrodites, our recommendation is that sex be assigned primarily on the basis of the external genitals and how well they lend themselves to surgical reconstruction in conformity with assigned sex, due allowance being made for a program of hormonal intervention. For older hermaphroditic infants, children and adults, our recommendation is that first consideration be given to the degree that a gender role has been ineradicably established in the sex already assigned, and that changes of sex be scrupulously avoided, except in rare and carefully appraised instances.

The chief objection to be raised against this second recommendation, as against the first, is that it also flagrantly disregards the possibility of depriving the group of nonsterile hermaphrodites of fertility in adult life. The answer to this objection is that actual child bearing as distinguished from potential biological fertility is not determined by chromosomal, hormonal, and gonadal sex alone. It is also determined by the social encounters and cultural transactions of mating and marrying, which are inextricably bound up with gender role and erotic orientation. Gender role may be established so thoroughly and irreversibly, despite chromosomal, gonadal or hormonal contradictions, that it cannot be modified in accordance with a change of sex by edict. Thus a boy, changed to wear dresses once ovaries were discovered, may continue to think, act and dream as the boy he was brought up to be, eventually falling in love as a boy, only to be considered homosexual and maladjusted by society. Alternatively, after the change, the gender role may be partly modified, but only at the cost of psychologic disorder and symptomatology sufficiently disabling to prevent marriage. In either case, the plan to preserve fertile gonads carries the seeds of its own defeat by ensuring that fertility never culminates in reproduction.

PSYCHOLOGIC MANAGEMENT: PARENTS

When hermaphroditism is recognized at birth and there is doubt about the baby's sex, this doubt should be frankly revealed to the parents. It is preferable that they remain in as much doubt about the sex of their child as they were before delivery than that they have to contradict a public announcement of a son or daughter. When finally the decision is made, the parents will be firm in the conviction that they have a son, or else a daughter, and unequivocal definiteness is to the child's subsequent advantage.

Ninety-nine times out of a hundred, the public construes an hermaphrodite as being half boy, half girl. The parents of an hermaphrodite should be disabused of this conception immediately. They should be given, instead, the concept that their child is a boy or a girl, one or the other, whose sex organs did not get completely differentiated or finished. A few simple embryological sketches (11, 12) showing the original hermaphroditism of all human embryos in the undifferentiated phase, and the late stage at which external genital similarity of males and females is still apparent, are of inestimable help in conveying this enlightening concept of genital unfinishedness (Figures 1 and 2). The average layman is totally ignorant of the fact that clitoris and penis, labia majora and scrotum, and labia minora and ventral skin of the penis are homologous structures (13). In fact, many women are ignorant of the existence of the clitoris on their own bodies. It comes as a surprise, therefore, to the parents of, say, a female pseudohermaphrodite that the "male organ" is really a clitoris which did not shrink to normal size. Acquaintance with proper genital terminology is also of great practical importance in enabling parents to explain their child's deformity to grandparents and other close relatives without such emotion-laden and embarrassing expressions as "morphodite" and "not being right down there." More important still, a proper vocabulary equips parents to talk straightforwardly with the child and to answer questions as the need arises.

A special problem of vocabulary arises when talking truthfully with parents, should a decision have been reached to allow a contradiction between gonadal structure and sex of rearing in their child—which should, for example, ordinarily be the decision when, despite testes, the phallus is clitoral sized. Under such circumstances, it is preferable to refer always to abnormally formed gonads or sex glands, and not to testes. An abnormally formed gonad may look testicular under the microscope, and one may tell the parents that this is so, but to say it in this way is totally different in its emotional effect from bluntly saying that the gonad is a testis. Medical people know that they call a gonad a testis despite tubular atrophy, absence of sertoli cells, and interstitial cells which produce estrogens, but lay people do not know all this.

Most parents need to be told that their child is not destined to grow up with

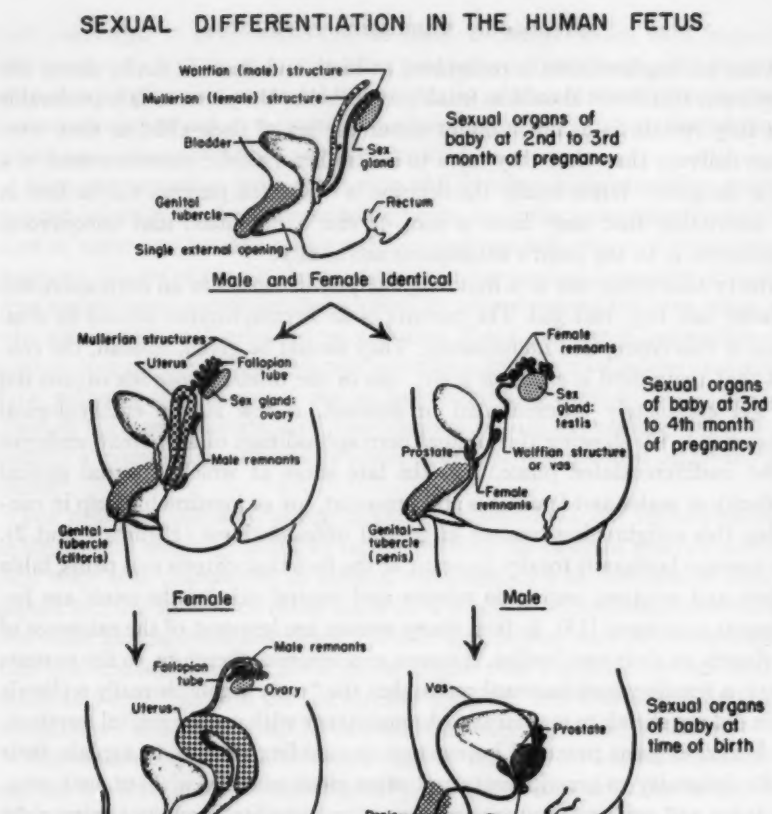

FIG. 1. Cross-section diagrams freely adapted from R. R. Greene and simplified for demonstrating to laymen the failure of complete sexual differentiation in hermaphroditism. (Drawn by Marie Vincent Kern)

abnormal and perverse sexual desires, for they get hermaphroditism and homosexuality hopelessly confused.

PSYCHOLOGIC MANAGEMENT: PATIENTS

The more manifest the signs of hermaphroditism, the more necessary are psychologic guidance and follow-up. As young as the age of two, hermaphroditic children begin to notice their difference from other boys or girls. Brought up as

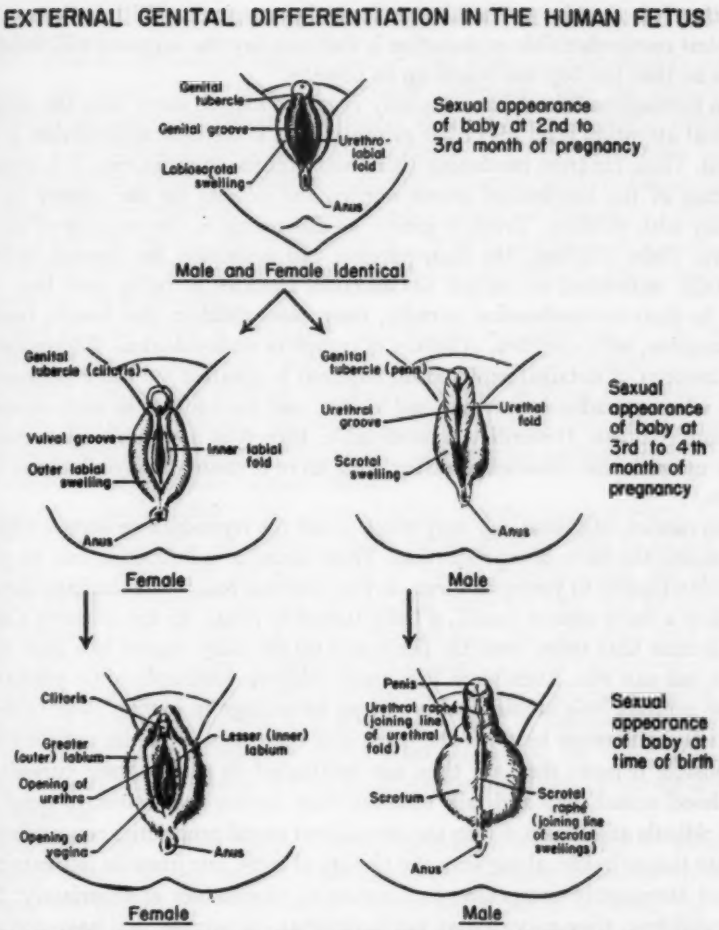

FIG. 2. Diagrams freely adapted from B. M. Patten and simplified for demonstrating to laymen the failure of complete external genital differentiation in hermaphroditism. (Drawn by Marie Vincent Kern)

a girl, a child with a big phallus may, putting two and two together, tell herself that she is part boy. But not invariably. If she has seen only baby girls naked, she may conjecture that all girls develop a phallus as they grow up or, contrarily, if she has seen only older girls naked, she may conclude that her phallus will diminish as she gets older. Sooner or later, however, she comes to realize her oddity. It is preferable that such a child know, from the time when she can first begin to comprehend it, that she has a clitoris like all other girls,

but that it is too big, and will be made smaller surgically. With little boys, the simplest comprehensible explanation is that one day the surgeons will finish the penis so that the boy can stand up to urinate.

An hermaphroditic child needs only eyes and ears to know that the focus of medical attention is on his or her genitals, even if the hermaphroditism is concealed. Thus, far from burdening them with unnecessary worries, it is actually a lifting of the burdens of secret worries and doubts for the doctor to talk frankly with children. Truth is seldom as distressing as the mystery of the unknown. Older children, like their parents, can assimilate the concept of being genitally unfinished to replace ill-conceived theories of being part boy, part girl. As their comprehension permits, these older children also benefit from an explanation, with sketches, of failure of complete embryological differentiation. The amount of detailed explanation required is greatest for those hermaphrodites whose paradox is external and visible, and least for those with concealed hermaphroditism. If sterility is predictable, then it is preferable for a child to grow up with this knowledge rather than have it dramatically disclosed later in life.

One cannot, of course, say very much about the reproductive system without explaining the facts of reproduction. These items of information can be given quite intelligibly to young children, as they become ready to assimilate them, in terms of a baby nest or pouch, a baby tunnel or chute, an egg without a shell, and sperms that swim from the penis and up the baby tunnel in a race to see which one can win. Even more than most children, hermaphroditic youngsters whose genitals look ambisexual are at an advantage in having their curiosity about the difference between the sexes and the origin of babies satisfied by a responsible person: they are then not motivated to satisfy their curiosity in childhood sexual play and talk whereby they expose themselves to neighborhood ridicule and gossip. Given the facts about moral proprieties concerning the private things in life, along with the biological facts, our juvenile patients have proved thoroughly competent in conducting themselves appropriately. Like other children, they may or may not masturbate in private, and have not been medically forbidden to do so, but they have presented no neighborhood problems of sexual conduct.

The most complex problems of psychologic management arise in those hermaphroditic children who are exposed to equivocating indecision and doubt on the part of parents and physicians and whose sex is debated or ridiculed by their contemporaries, so that they do not establish their gender with unambiguous certainty. These ambiguously uncertain children include those subjected to change of sex after the early infancy period. The problems and time involved in handling these cases require the services of a specialist in psychiatric medicine.

PSYCHOLOGIC MANAGEMENT: SURGERY

As a general psychologic rule, the earlier that signs of ambisexuality are corrected, the better, both from the parents' viewpoint and the child's. But operative decisions are always contingent on estimates of surgical safety and success. Concerning surgical safety, for example, it is risky for female pseudohermaphrodites whose hyperadrenocorticism is complicated by sodium loss to undergo surgery before salt retention has been firmly established. Concerning surgical success, except for clitoridectomy, greatest success with genital repair and reconstruction is usually obtained after the organs have grown and are of a good size to work with.

When surgical delay is indicated, children can accommodate to the impositions and embarrassments of a genital anomaly, but it is imperative that they be told about prospective medical and surgical plans. Without this information, they are likely to construe that they are freaks. With it, they can discuss and elaborate tactics for dealing with interim emergencies such as, in the case of hypospadiac boys, school urinary habits.

Not only should children be informed of prospective surgical plans, but also they should invariably be given a simple explanation of what to expect of an operation. A three year old girl about to be clitoridectomized, for example, should be well informed that the doctors will make her look like all other girls and women. Girls should also know, incidentally, that whereas boys have a penis, girls have a vagina—in juvenile vocabulary, a baby tunnel—as a double insurance against childish theories of surgical mutilation and maiming. It is also useful to inform children, as a matter of course, of preoperative and anesthetic routine and of what to expect postoperatively, for these apparently trivial details are the source of their most intense anticipatory terrors and misconceptions.

Clitoridectomy is the genital surgical procedure most likely to rouse psychologic debate among experts, particularly on the issue of loss of erotic sensation. We have sought information about erotic sensation from the dozen nonjuvenile hyperadrenocortical women we have studied. There has been no evidence of a deleterious effect of clitoridectomy. None of the women experienced in genital practices reported a loss of orgasm after clitoridectomy. All of the patients were unanimous in expressing intense satisfaction at having a feminine genital morphology after the operation. None of them had been antagonistic to the plan for clitoridectomy. Each had regarded herself as a woman and had established a feminine gender role. It would be most unwise to dictate clitoridectomy to a gonadal female who had established a masculine gender role and considered himself a boy or man. Formerly, complete clitoral extirpation was required in some hyperadrenocortical females who had painful priapism of the amputated stump, but cortisone abolishes priapism so that amputation is

nowadays sufficient. Though there is considerable evidence that an amputated clitoris is erotically sensitive, enough uncertainty remains to require conservatism in recommending clitoridectomy for hermaphrodites living as women in whom there is neither a blind vaginal pouch nor a vagina opening into a urogenital sinus.

When plastic reconstruction of the vagina is delayed until postadolescence, the patient should be made to feel free to request the operation whenever it suits her, and not only after she is engaged or married. It is unnecessarily embarrassing for a woman to have to explain and confess her vaginal shortcomings to a prospective husband.

CASE ILLUSTRATION

The following case report has been chosen to illustrate the type of case in which the pros and cons of a change of sex provoke considerable debate. The final decision in this instance was against a change of sex, chiefly on psychologic grounds which are spelled out and briefly documented in the report.

Introductory data: A diagnosis of hyperadrenocortical female pseudohermaphroditism was first made when the patient was three years and seven months old, at which time he was referred for psychologic appraisal. He had normal female internal reproductive structures, except that the vagina opened into the urethra near the bladder. He had already begun to virilize precociously in the typical hyperadrenocortical fashion. He had always lived as a boy. When he was born, the mother said, she was told that the sex of her baby was uncertain, but by the third day she was informed unequivocally that she had a son.

From the age of a year onward, the child had been hospitalized elsewhere, at approximately six-monthly intervals, for four successive stages of hypospadiac repair. The repair had not yet been completed and the child was still obliged to sit to urinate. The surgical consultant reported that further plastic surgery could be carried out to provide an adequately functioning penis, provided the mullerian system were removed, mucosa from the hidden vagina being used to fashion a penile urethra. The child could also be surgically feminized, with amputation of the phallus and construction of labia majora from the very rugose skin of what looked like an empty scrotum. Subsequent hormonal feminization could then have been induced with cortisone.

Family: The mother combined big-city flair and quick wits with the simple and artless folkways of a Scandinavian upbringing and meager education. She appeared to be a sincere, candid woman and mother, visibly harassed by the arduousness of synchronizing her son's hospitalization with all the other chores and duties of caring for her family. She gave the impression of not daring to give vent to her feelings and worries about her son lest, having let go, she find herself unable to resume mastery of herself again and carry on with her other responsibilities. Balancing the family budget was a constant responsibility, for her husband had been partially incapacitated for fifteen years by an inability to maintain postural equilibrium.

The family included five children: Bill, 19; David, 16; Bob, 7; Norman, the patient, 3½; Kate, the only girl, 1 year. There had been no other pregnancies.

The mother gave no sign of knowing that fresh doubts about the sex of her fourth child had arisen, and that the question of a change of sex had been asked. She had wondered initially when the child was a baby whether he were part girl. She had even wondered

whether to give credence to her husband's conjecture that her intense longing for a daughter instead of a fourth son had caused the baby's genital ambiguity. Subsequently she had not for one moment conceived the child to be the daughter she had so much longed for. It appeared rather that she had made restitution for even a conjectured prenatal influence by accepting with redoubled certainty the maleness of the child and rearing him accordingly.

First impression of child: "As soon as he recognized my face as unfamiliar, he approached me, saying over and over again: 'Got to call my Mommy.' There was a look of stark terror about him, and a note of frantic urgency in his voice. He did not object to a genital examination, but kept perseverating, uneasily: 'The nurse cut my wee-wee.' I could not find much logical coherence between this and other reiterated sentences, and could not understand some of his baby-talk pronunciations. I was left wondering whether the child had some kind of cerebral defect, or whether he was simply in the midst of a hospital admission panic."

Intelligence test: The Revised Stanford-Binet Intelligence Scale was administered. Chronological Age three years and seven months; Mental Age two years and eleven months; IQ 81—dull normal level. The child was very friendly and cooperative. It was judged he did the best he was capable of on the test, but his general uneasiness at being in a hospital may well have had an adverse effect on his IQ score.

Interviews with child: The child was seen by each of the three investigators. Two of eight hours of conversation were recorded. He made an extremely cordial friendship with the man who saw him most frequently. With women he was slow to get friendly and at ease, which seemed to have some connection with his misconception that nurses, and not doctors, cut his penis as well as managing the postoperative procedures which hurt him.

With familiarity, the child's speech became easier to understand. It was rather more difficult than is usual with three and a half year olds to engage in logically coherent dialogue. Though his statements were not always linked coherently together and were sometimes childishly inconsistent, it was possible to trace a thread of continuity.

He came to the hospital, he said, because his mother brought him in the choo-choo train. And he also came to the hospital because: "The nurse cut on my wee-wee. The nurse hurt me. Cut on my wee-wee." Immediately he went on, reiterating: "I got to call my Mommy," as he tried to reach the telephone on the desk. "Got to call my Mommy. Take me in the choo-choo train. Home." By implication, he wanted to get out of the hospital before there was any more cutting on his wee-wee. Spontaneously he continued: "The nurse cut on Bobo's wee-wee" (Bob, his seven year old brother). It was not clear whether this was a statement of fact signifying that Bob's penis was small like his own, while the older brothers had large ones, or whether it was an expectation that Bob would be treated like himself. Kate, the sister, was mentioned next. She had no wee-wee. The nurse had cut it and taken it away. "Billy (the oldest brother) will bring Katie a new wee-wee," and other presents too. This new kind of organ was called a "koll wee-wee." It was the sort that Billy himself had. "Billy will bring me a koll wee-wee, too. Billy is nice to me."

Subsequently it was learned that the patient had spontaneously asked his oldest brother, Billy, away from home in the service, for "a new wee-wee," while talking on long distance with him. On the ward, the child was also spontaneous with impromptu talk about his wee-wee and its being cut, in conversation with the nursing and house staff. For all of his excessive penis-consciousness, he was not self-conscious in a prudish, embarrassed sense.

Like many infants, the child got muddled about the relationships of brother and sister. But he also got muddled about who were boys and who was girl in his family, which is unusual at three and a half. As for himself, by contrast, there was absolutely no question: he always answered definitively that he was a boy. In point of fact, to the many people who asked him his name, his reply was the single monosyllable: "Boy." He gave the name of his

boy-dressed doll in the same way. Once when pressed for the doll's proper name, he tried to satisfy with: "Boy doll-girl," but there was never any such ambiguity about himself. Under similar pressure for his own proper name, he gave his age instead, saying two, one, three, with fingers held up, not quite sure of himself. Despite all this precision about being a boy at the cost of indecision about his name, he quickly agreed that his name was Norman when asked if it was.

In his general manner and bearing, the child was quite unlike little girls of his age and completely like other little boys. The mother remarked on how boyish she found him—exactly as boyish as her three other sons had been. The house staff and nurses were unanimous that their impulsive feelings and responses to the child were as to other boys, not as to a girl nor an undecided misfit. Other children on the ward and in the playroom accepted him unhesitatingly as a boy. After his initial panicky reaction, he acted boisterously and full of boyish high spirits, without being seclusive, timid or inhibited. So far as could be judged, he was much relieved when informed that there would be no surgery during this admission, but that when he got bigger his penis would be, not taken away, but finished so that he could stand to urinate.

PSYCHOLOGIC APPRAISAL

No one who had any sustained contact with this child could fail to observe that he was extraordinarily cognizant of his penis, even for an hypospadiac child, and alarmed by the history and prospects of its surgical alteration. In a typically childish way, he had grossly misconstrued his surgical experiences to signify that his penis was being mutilated and perhaps might suffer the fate he conceived had befallen his sister's genitals. His fantasied remedy for his plight was that his oldest brother would buy him a new and perfect penis. In an older person, this kind of reiterated illogical thinking would be identified as delusional and psychopathological, though it is regarded as benign in a child so young. It is not so completely benign, however, that one can affort to treat it casually and with indifference. The longer such misconceptions stay unrectified, they become increasingly ineradicable as a source around which more and more allied or derivative misconceptions cumulate. As this cumulative distortion of thinking and judgment progresses, there is reciprocal distortion of the balance between action and inhibition, between active meeting and mastering of life's challenges and control and discipline of actions and conduct, until a condition of frank psychopathological disorder is in evidence. At the age of three and a half, this child showed no signs of such disorder—only the signs that he was a good candidate in whom disorder might develop.

A very good way to promote development of psychopathological symptoms in this child would have been to subject him to a fifth genital operation to amputate the four-times-repaired hypospadiac phallus. So far as a change of sex for such a child is concerned, one is, therefore, on the horns of a dilemma. On the one hand, to amputate the phallus without delay would magnify all the child's misconceptions and prove that they had been only too well founded, without in any way proving to him his feminity. On the other hand, delay of

surgical feminization would leave strong genital evidence of maleness and impede assimilation and development of a new, feminine gender role and orientation.

The only conceivable way of making a change of sex and also of resolving this dilemma would be through an intensive program of psychiatric follow-up, both with the child and, to a lesser degree, with the members of his family, after the family had moved to an entirely new neighborhood. As a practical matter, this strategy was out of the question, for the family was impecunious and there was no other source of funds to meet all of the very costly exigencies.

For this child, the risk of a change of sex ending in psychiatric disaster was judged too great to justify the change. The greater medical wisdom lay in planning for a sterile man to be physically and mentally healthy, and efficient as a human being, than for a probably fertile woman to be physically well but psychologically a misfit and a failure as a woman, a wife, or a mother. Accordingly, it was planned to prevent precocious epiphysial fusion and too rapid advance of secondary sexual virilization by treatment with cortisone until the appearance of beginning feminization, and then to perform an ovariectomy and to discontinue cortisone. Completion of hypospadiac repair was postponed until the child was older and less psychologically menaced by the procedure.

SUMMARY

There are seven sexual variables which may operate independently of one another in hermaphroditism. They are: chromosomal sex; gonadal sex; hormonal sex; external genital morphology; accessory internal genital morphology; assigned sex and rearing; and gender role, including psychosexual orientation, which becomes built-up through experiences encountered and transacted. On the basis of chromosomal, gonadal and hormonal sex, seven varieties of hermaphroditism have been described, but these three criteria, singly or together, are never sufficient when practical decisions about assignment or change of sex have to be made. In the case of neonatal and very young infant hermaphrodites, we recommend that sex be assigned primarily, though not exclusively, on the basis of the external genitals and how well they lend themselves to surgical reconstruction in conformity with assigned sex, due allowance being made for a program of hormonal intervention, if indicated. For older hermaphroditic infants, children and adults, we recommend that first consideration be given to the degree that a gender role has been indelibly established in the sex already assigned, and that changes of sex be scrupulously avoided, except in rare and carefully appraised instances, in order to avoid hazardous psychiatric sequelae. Judged in conjunction with each of the seven clinically recognizable varieties of hermaphroditism, these recommendations make eminent practical sense. A case is reported to illustrate the methods of studying attitudes and diagnosing gender role and applying these concepts in practical decisions. The

psychologic management of parents of hermaphrodites and of patients themselves, with special reference to genital surgery, derives in its specific details from a general policy of frank and straightforward discussion and explanation. A clear and explicit understanding of basic principles is a prerequisite to frank and straightforward discussion.

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AN EXAMINATION OF SOME BASIC SEXUAL CONCEPTS: THE EVIDENCE OF HUMAN HERMAPHRODITISM*

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INTRODUCTION

Despite advancements of knowledge in embryology and endocrinology, most people have continued to make an absolute dichotomy between male and female—a dichotomy as seemingly axiomatic as the distinction of day from night, black from white. In psychology and psychiatry, this dichotomy is represented in the conception of predominant masculinity or feminity of the sexual instinct or drive.

A comprehensive theory of instinctive sexuality was first expounded to a medical audience by Freud at the beginning of the present century (1). Developing and expanding earlier instinctive theories, Freud utilized, inter alia, the conception of innate bisexuality, namely, that instinctive masculinity and instinctive femininity are present in all members of the human species, but in differing proportions.

Freud construed his theory of innate and constitutional psychic bisexuality on the basis of embryological evidence of an hermaphroditic phase in human embryonic differentiation, and on the basis of anatomical evidence in congenital hermaphroditism itself. At a later date, he adduced the evidence of early experiments in endocrinology concerning hormonal reversal of sex in animals as a further support for his bisexual theory.

In an endeavor to ascertain if new and additional information relevant to psychologic theory of sexuality might be obtained from the study of hermaphroditism, the authors have, for the past four years, systematically been making psychologic studies of hermaphroditic patients. The work has been done in close cooperation with Dr. Lawson Wilkins who has been responsible for diagnostic, endocrine and other medical studies of the majority of the patients.

The sexual incongruities which occur in hermaphroditism involve divers con-

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tradictions, singly or in combination, between six variables of sex. These six variables are:

1. Assigned sex and sex of rearing;
2. External genital morphology;
3. Internal accessory reproductive structures;
4. Hormonal sex and secondary sexual characteristics;
5. Gonadal sex;
6. Chromosomal sex.

Patients showing various combinations and permutations of these six sexual variables may be appraised with respect to a seventh variable:

7. Gender role and orientation as male or female, established while growing up*.

Thus one is enabled to ascertain something of the relative importance of each of the six variables in relation to the seventh.

Patients in whom ambisexual contradictions exist include seven subgroups in the traditional diagnostic category of hermaphroditism (2, 3), and a group traditionally diagnosed as ovarian agenesis but now more accurately named gonadal agenesis, or dysgenesis, since the chromosomal pattern is male (4, 5) (Table I). We have studied 76 patients in all.

CHROMOSOMAL SEX

Barr's technique of chromosomal sex determination from skin biopsies has, since January 1954, revealed the chromosomal sex of some of our patients (7, 8)†. Though it is too early to make a final definitive statement, it appears reasonably likely that, among the eight varieties of ambisexual development, a female chromosomal pattern is always present in varieties 1 and 2, and a male chromosomal pattern in varieties 6, 7, and 8. It happens that eight true hermaphrodites so far reported on have had a female, and four a male chromosomal pattern. The chromosomal sex of males in whom mullerian differentiation is relatively unarrested has yet to be elucidated. Gonadal agenesis (dysgenesis) may occur either in chromosomal males or chromosomal females who, in all other respects, are identically feminine.

In the interests of precision, only those patients from whom skin biopsies

* By the term, gender role, we mean all those things that a person says or does to disclose himself or herself as having the status of boy or man, girl or woman, respectively. It includes, but is not restricted to sexuality in the sense of eroticism. Gender role is appraised in relation to the following: general mannerisms, deportment and demeanor; play preferences and recreational interests; spontaneous topics of talk in unprompted conversation and casual comment; content of dreams, daydreams and fantasies; replies to oblique inquiries and projective tests; evidence of erotic practices and, finally, the person's own replies to direct inquiry.

† Chromosomal sex determinations were done by Dr. M. L. Barr and Dr. K. L. Moore at the University of Western Ontario.

TABLE I

*Varieties of Somatic Ambisexual Development**

1. Congenitally hyperadrenocortical female	42
2. Female, with well differentiated phallus, normal ovaries and normal internal reproductive structures	2
3. True hermaphroditism	1
4. Male, with unarrested mullerian differentiation:	
(a) with normal penis and one or both testes cryptorchid	0
(b) hypospadiac and cryptorchid	3
5. Gonadal agenesis (dysgenesis), with simulant female infantile body morphology and male chromosomes	11
6. Simulant female, with testes, blind vagina, mullerian vestiges and breasts	3
7. Cryptorchid male hypospadiac, with breasts at puberty:	
(a) with urogenital sinus	1
(b) with blind vaginal pouch	0
8. Cryptorchid male hypospadiac:	
(a) with urogenital sinus	11
(b) with blind vaginal pouch	2
	76

* For the purposes of this classification, the criterion of male and female is gonadal. The classification is based on the study of over 300 cases in the literature, in English, of the last half century (6). Within each category, patients resemble one another closely enough to be strictly comparable as somatic units for the purpose of psychologic study.

TABLE II

CHROMOSOMES AND REARING CONTRADICTORY
19 CASES

CHROMOSOMES	GONADS	ENDOGENOUS HORMONAL SEX	INTERNAL ACCESSORY ORGANS	EXTERNAL GENITAL MORPHOLOGY	ASSIGNED SEX AND REARING	GENDER ROLE	TYPE OF AMBISEXUAL DEVELOPMENT
♂	none	none	♀	♀	11 ♀	11 ♀	gonadal agenesis (dysgenesis)
♂	♂	1 ♀ 2 [illegible]	vestigial	♀	3 ♀	3 ♀	simulant female
♂	♂	1 ♂ 3 [illegible]	vestigial	♀	4 ♀	4 ♀	cryptorchid male hypospadiac
♀	♀ left ♂ right	♀	♀	♀	1 ♂	1 ♂	true hermaphroditism

have been taken, and the chromosomal sex actually determined, are included in Table II. The 19 cases listed in this table are those in which there was a contradiction between chromosomal sex and the sex of assignment and rearing. In every instance, the person established a gender role and orientation con-

TABLE III

GONADS AND REARING CONTRADICTORY
20 CASES

GONADS	CHROMOSOMES	ENDOGENOUS HORMONAL SEX	INTERNAL ACCESSORY ORGANS	EXTERNAL GENITAL MORPHOLOGY	ASSIGNED SEX AND REARING	GENDER ROLE	TYPE OF AMBISEXUAL DEVELOPMENT
♀	♀	♀	♀	♂	2 ♂	2 ♂	female with phallus and ovogenesis
♀	♀	♂	♀	♂	4 ♂	4 ♂	hyperadrenocortical female
♂	?	1 ♂ 2 juv.	♂	♂	3 ♀	3 ♀	male with uncrested mullerian differentiation
♂	♂	1 ♀ 2 juv.	vestigial	♀	3 ♀	3 ♀	simulant female
♂	♂	4 ♂ 4 juv.	vestigial	♂	8 ♀	5 ♀ 1 ♀→♂ 2 ♂	cryptorchid male hypospadiac

sistent with assigned sex and rearing, and inconsistent with chromosomal sex. Thus, it is convincingly clear that gender role and orientation as male or female evidenced itself independently of chromosomal sex, but in close conformity with assigned sex and rearing.

GONADAL SEX

Among the 76 patients, there were 20 in whom a contradiction was found between gonadal sex and the sex of assignment and rearing (Table III). All but 3 of these 20 disclosed themselves in a gender role fully concordant with their rearing. Gonadal structure per se proved a most unreliable prognosticator of a person's gender role and orientation as man or woman, boy or girl. By contrast, assigned sex and rearing proved a most reliable one.

HORMONAL SEX

To consider now the relationship between hormonal sex and gender role: hormonal sex must be distinguished from gonadal structure, for ovaries do not always make estrogens, nor testicles androgens. The ovaries of hyperadrenocortical female pseudohermaphrodites are inert, and though their adrenals produce an excess of estrogens as well as of androgens, androgenic activity dominates and the body is excessively virilized. The testes of male pseudohermaphrodites of the simulant female variety produce estrogens which feminize the body.

Table IV summarizes the data on the 27 patients who went through and

TABLE IV

HORMONAL SEX AND REARING CONTRADICTORY

27 POSTPUBERTAL CASES

ENDOGENOUS HORMONAL SEX	CHROMOSOMES	GONADS	INTERNAL ACCESSORY ORGANS	EXTERNAL GENITAL MORPHOLOGY	ASSIGNED SEX AND REARING	GENDER ROLE	TYPE OF AMBISEXUAL DEVELOPMENT
♀	♀	♀	♀	⚥	2 ♂	2 ♂	female with phallus and ovogenesis
♀	♂	♂	vestigial	⚥	1 ♂	1 ♂	cryptorchid male hypospadiac with breasts
⚥	♀	⚥	⚥	⚥	1 ♂	1 ♂	true hermaphroditism
♂	♀	♀	♀	⚥	18 ♀	17 ♀ 1 ⚥	hyperadrenocortical female
♂	?	♂	⚥	⚥	1 ♀	1 ♀	male with unarrested mullerian differentiation
♂	♂	♂	vestigial	⚥	4 ♀	1 ♀ 1 ♀→♂ 2 ⚥	cryptorchid male hypospadiac

beyond a puberty—or, in the case of hyperadrenocorticism, a precocious puberty-equivalent—in which hormonal influences produced a secondary sexual development contradictory of the sex in which they were living. Subsequently the contradiction was corrected with hormonal therapy and, where indicated, with plastic surgery. Psychologic data were gathered before treatment, when possible, otherwise retrospectively.

Of the 27 people whose hormonal functioning and secondary sexual body morphology contradicted their assigned sex and rearing, only 4 became ambivalent with respect to gender role as male or female. All four had been reared as girls. One, acting on his own initiative, began living as a man from the age of sixteen onward. The other three, while living as women, showed some degree of bisexual inclination. These four patients do not, in themselves, offer any convincing evidence of hormonal sex as a causal agent in the establishment of maleness or femaleness of gender role: the patient who lived as a man declined testosterone substitution treatment after surgical castration for malignancy, and the three who lived as women had been thoroughly feminized on estrogen substitution treatment. Moreover, the other 23 of the 27 patients established a gender role consistent with their assigned sex and rearing, despite the embarrassment and worry occasioned by hormonal contradictions. Like gonadal sex, hormonal sex per se proved a most unreliable prognosticator of a person's gender role and orientation as man or woman, boy or girl.

TABLE V

INTERNAL ACCESSORY ORGANS AND REARING CONTRADICTORY

18 CASES

INTERNAL ACCESSORY ORGANS	CHROMOSOMES	GONADS	ENDOGENOUS HORMONAL SEX	EXTERNAL GENITAL MORPHOLOGY	ASSIGNED SEX AND REARING	GENDER ROLE	TYPE OF AMBISEXUAL DEVELOPMENT
♀>♂	♀	♀	♀	♀	2 ♂	2 ♂	female with phallus and ovogenesis
♀>♂	♀	♀	♂	♀	4 ♂	4 ♂	hyperadrenocortical female
♂>♀ vestigial	♂	♂	1 ♀ 2 ♂	♀	3 ♀	3 ♀	simulant female
♂>♀ vestigial	♂	♂	4 ♂ 4 ♂	♂	8 ♀	5 ♀ ♂ 1 ♀ ♂ 2 ♂ ♀	cryptorchid male hypospadiac
♂♀	♀	♂♀	♂♀	♂♀	1 ♂	1 ♂	true hermaphroditism

INTERNAL ACCESSORY ORGANS

Since the uterus is the organ of menstruation, and the prostate the major organ of seminal fluid secretion, it is necessary to compare maleness or femaleness of gender role with internal reproductive equipment. There were 17 cases in whom assigned sex and rearing was inconsistent with predominant male or female structures internally (Table V). Gender role agreed with rearing in 14 of these 17. The 3 remaining were the same three individuals as deviated in Tables III and IV.

In estimating the significance of the comparison in Table V, one must bear in mind that only rarely in hermaphroditism does either the uterus or the prostate reach full functional maturity, without medical intervention. Of the 17 cases, there were only 3 for whom this statement did not hold: though all three had a functional uterus, they had been reared as boys and had a thoroughly masculine gender role and outlook. So far as the evidence goes, there is no reason to suspect a correlation between internal accessory organs and maleness or femaleness of gender role.

EXTERNAL GENITAL APPEARANCE

It goes without saying that the external genitals are the sign from which parents and others take their cue in assigning a sexual status to a neonate and in rearing him thereafter, and the sign, above all others, which gives a growing child assuredness of his or her gender. Nonetheless, it is possible for an hermaph-

TABLE VI

EXTERNAL GENITAL APPEARANCE AND REARING CONTRADICTORY

23 CASES

PREDOMINANT EXT. GENITAL APPEARANCE	CHROMOSOMES	GONADS	ENDOGENOUS HORMONAL SEX	INTERNAL ACCESSORY ORGANS	ASSIGNED SEX AND REARING	GENDER ROLE	TYPE OF AMBISEXUAL DEVELOPMENT
♂	♀	♀	♂	♀	18 ♀	18 ♀	hyperadrenocortical female
♂	?	♂	1 ♂ 1 ♂	♀	2 ♀	2 ♀	male with unarrested mullerian differentiation
2 ♀ 3 ♂	2 ♂ 3 ♂	2 ♂ 3 ♂	2 ♂ 2 ♂ 1 ♂	2 vestigial 3 vestigial	2 ♂ 3 ♀	2 ♂ 1 ♀→♂ 2 ♀	cryptorchid male hypospadiac
♀	♂	♂	♀	vestigial	1 ♂	1 ♂	cryptorchid male hypospadiac with breasts

rodite to establish a gender role fully concordant with assigned sex and rearing, despite a paradoxical appearance of the external genitals.

There were 23 among our 76 patients who, at the time they were studied, had lived for more than two-thirds of their lives with a contradiction between external genital morphology and assigned sex (Table VI). For one reason or another, they did not receive surgical correction of their genital deformity in infancy, but lived with a contradictory genital appearance for at least five and for as many as forty-seven years. In all but one instance, the person had succeeded in coming to terms with his, or her anomaly, and had a gender role and orientation wholly consistent with assigned sex and rearing.

It is not contended that these people encountered no difficulties in their lives. On the contrary, there was considerable evidence that visible genital anomalies occasioned much anguish and distress. Distress was greatest in those patients whose external genital morphology flagrantly contradicted, without hope of surgical correction, the sex in which they had grown up and established, indelibly, their gender role and orientation as boy or girl, man or woman. Distress was also quite marked in patients who had been left in perplexed confusion about the sex to which they belonged, in consequence either of parental or medical indecision, or of insinuations from age-mates that they were half boy, half girl. Uniformly, the patients were psychologically benefited by corrective plastic surgery, when it was possible, to rehabilitate them in the sex of assignment and rearing. Only one patient failed to take advantage of plastic surgery. Instructively enough, he was the person who, on his own initiative, changed his birth certificate and began living as a man from the age of sixteen onward. He was unable to summon up enough courage to have his genitals masculinized.

It is relevant to note in passing that psychotic symptoms in all of the patients were conspicuous by their absence. In remarkably few instances, evidence of neurotic symptomatology was apparent. In some, but by no means all patients, feelings of bashfulness, shame and oddity were to the fore, and they had great initial diffidence in talking about themselves.

ASSIGNED SEX AND REARING

Chromosomal sex, gonadal sex, hormonal sex, internal accessory reproductive organs and external genital morphology—each of these five variables of sex has passed successively in review and has been compared first with assigned sex and rearing and, second, with the gender role and orientation as boy or girl, man or woman, which the person established while growing up. In only 4 cases among 76 was any inconsistency between rearing and gender role observed, despite the many inconsistencies between these two and the other five variables of sex.

Evidently there is a very close connection between, on the one hand, the sex to which an individual is assigned, and thenceforth reassigned in a myriad subtle ways in the course of being reared day by day, and, on the other hand, the establishment of gender role and orientation as male or female.

GENDER ROLE AND ORIENTATION

In the light of hermaphroditic evidence, it is no longer possible to attribute psychologic maleness or femaleness to chromosomal, gonadal or hormonal origins, nor to morphological sex differences of either the internal accessory reproductive organs or the external genitalia. Conceivably, of course, instinctive masculinity or feminity may be attributed to some other innate bodily origin. For example, Krafft-Ebing (9) among others has suggested special brain centers. There is, however, no support for such a conjecture when, as may happen in hermaphroditism, among individuals of identical diagnosis, some have been reared as boys, some as girls (Table VII).

There are 55 cases represented in Table VII, including the only 4 in the whole series of 76 in whom ambivalence of gender role was found. The four had been reared as girls. Among the 51 remaining, 42 reared as girls had established a feminine gender role and orientation, while 9 reared as boys had established a masculine gender role and orientation.

From the sum total of hermaphroditic evidence, the conclusion that emerges is that sexual behavior and orientation as male or female does not have an innate, instinctive basis.

In place of a theory of instinctive masculinity or femininity which is innate, the evidence of hermaphroditism lends support to a conception that, psychologically, sexuality is undifferentiated at birth and that it becomes differentiated as masculine or feminine in the course of the various experiences of growing up.

TABLE VII

MALE AND FEMALE ASSIGNED SEX IN PATIENTS WITH SAME DIAGNOSIS

55 CASES

ASSIGNED SEX AND REARING	GENDER ROLE	TYPE OF AMBISEXUAL DEVELOPMENT
38 ♀ 4 ♂	37♀, 1♂ 4♂	hyperadrenocortical female
8 ♀ 5 ♂	5♀,1♀→♂,2♂ 5♂	cryptorchid male hypospadiac

Those who find the concept of instinct or drive congenial may choose to say that there is a sexual instinct or drive that is undifferentiated and genderless at birth. In that case, sexual drive is neither male nor female to begin with, and it can be assumed to have no other somatic anchorage than in the erotically sensitive areas of the body. So limited, sexual drive becomes a special example of a kinaesthetic or haptic drive—an urge to touch and be touched, an urge for bodily contact.

Those who find the concept of drive uncongenial may choose simply to say that in the human species there are erotically sensitive areas of the body, especially the genital organs, and that these areas are sometimes stimulated and used by oneself or another person. In the course of growing up, a person's sexual organ sensations become associated with a gender role and orientation as male or female which becomes established through innumerable experiences encountered and transacted.

Our studies of hermaphroditism have pointed very strongly to the significance of life experiences encountered and transacted in the establishment of gender role and orientation. This statement is not an endorsement of a simple-minded theory of social and environmental determinism. Experiences are transacted as well as encountered—conjunction of the two terms is imperative—and encounters do not automatically dictate predictable transactions. There is ample place for novelty and the unexpected in cerebral and cognitional processes in human beings.

Novelty and unexpectedness notwithstanding, cerebral and cognitional processes are not infinitely modifiable. The observation that gender role is established in the course of growing up should not lead one to the hasty conclusion that gender role is easily modifiable. Quite the contrary! The evidence from examples of change or reassignment of sex in hermaphroditism, not to be presented here in detail, indicates that gender role becomes not only estab-

lished, but also indelibly imprinted. Though gender imprinting begins by the first birthday, the critical period is reached by about the age of eighteen months. By the age of two and one-half years, gender role is already well established.

One may liken the establishment of a gender role through encounters and transactions to the establishment of a native language. Once imprinted, a person's native language may fall into disuse and be supplanted by another, but is never entirely eradicated. So also a gender role may be changed or, resembling native bilingualism, may be ambiguous, but it may also become so indelibly engraved that not even flagrant contradictions of body functioning and morphology may displace it.

CASE ILLUSTRATION

The following illustrative case has been chosen because it shows convincingly how gender role and orientation may be fully concordant with the sex of assignment and rearing, despite extreme contradiction of the other five variables of sex.

Introductory data: The patient was twenty-four years old and married at the time of psychologic study. He had lived all of his life as a male. Except for a small hypospadiac phallus and fused, empty labioscrotum, he was found to be anatomically and physiologically female when, at the age of eleven and one-half years, he entered a hospital because his breasts had begun to enlarge and his body had grown increasingly feminine in contour. His genital abnormality, of which there was no known familial incidence, had been known to exist since birth. The penis had not enlarged significantly with the onset of puberty. Its glans was completely hidden by wrinkles of foreskin. Although the urethral orifice was located at the base instead of the tip of the penile shaft, the boy was able to stand to urinate. Testicles could not be palpated. The scrotum was contracted and not in the least pendulous.

Exploratory laparotomy revealed a uterus, fallopian tubes, bilateral parovarian cysts, and two cystic gonads in the position of ovaries. The uterus did not connect with a normal vagina, but appeared to open into the upper urethra. No organs of the male reproductive system were discerned internally. The uterus, tubes and gonads were removed, especially as the parents thought their child should remain a boy. Only two sections of the gonads were examined microscopically. They revealed ovarian structure, with primordial follicles containing ova, a few intermediate forms of developing graafian follicles, and several large cyst-like structures lined with granulosa cells.

Postoperatively, male hormone therapy was instituted and continued regularly thereafter. When the patient was twenty-one, therapy was withheld for about nine months on a trial basis, with a result that he felt weak and easily fatigued. Two years later, in preparation for marriage, he underwent plastic surgery in order to transpose and straighten the penis. At this time he appeared quite masculine in stature, though the breasts had remained slightly enlarged. Facial hair required shaving every second or third day. The pubic hair was masculine in distribution and the voice deep. A small, soft prostate was palpable.

First impression: "At first sight and throughout the first meeting with this man, I kept thinking that nothing in his general appearance, manner or conversation betrayed a single hint of the information filed away in his medical record. He would pass anywhere as the advanced graduate student that he was. So much was he a young man, indeed, that I wondered briefly if it might be best to leave well enough alone and not risk stirring up doubts and fore-

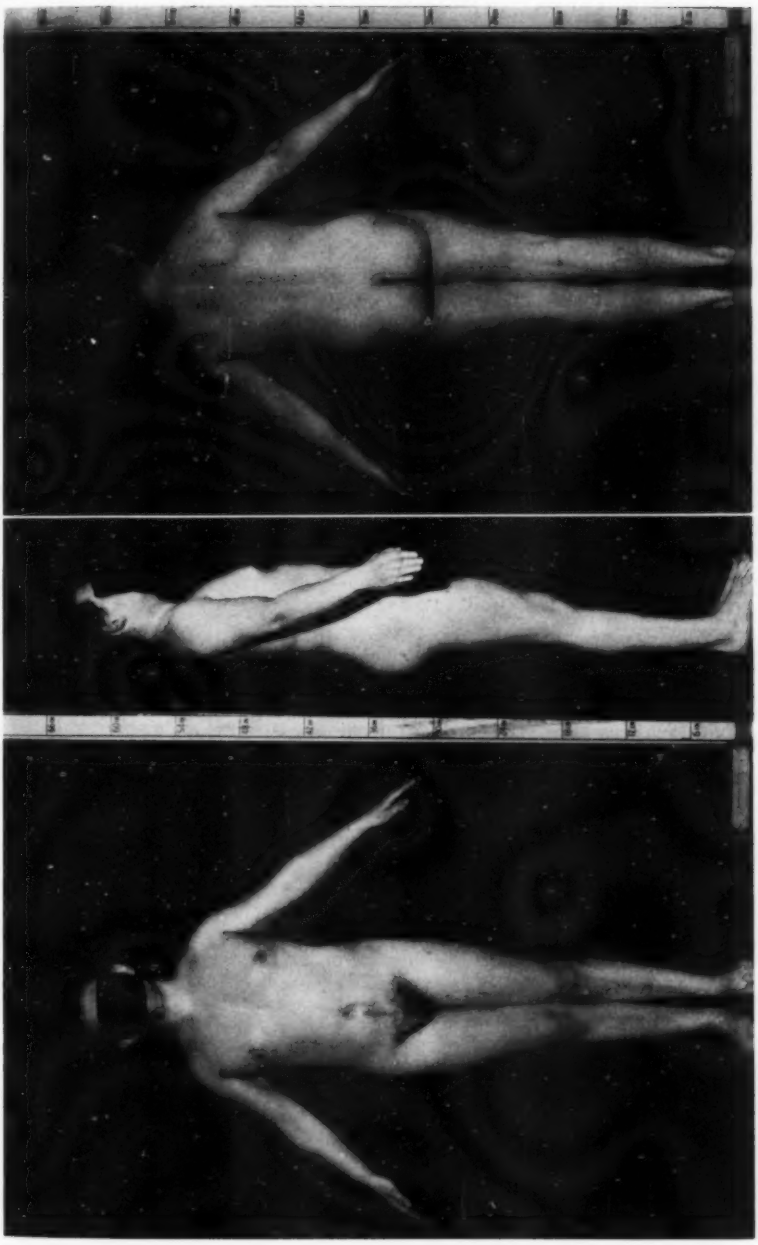

FIG. 1

bodings. He quickly grasped the purpose of a psychological research study, however, and readily consented to cooperate."

Interviews: At all times the man talked fluently and spontaneously, unperturbed by recording apparatus, with no signs of withholding information through embarrassment. He had been told that utmost discretion would be used in concealing his identity—which was an imperative expediency in view of his educational affiliations and associations—and that he would be able to censor the manuscript of the case report before its circulation or publication. Most of the interviews and tests were completed within a single week, with only one examiner, over a period of about fifteen hours. After that there were a few meetings, including two or three informal visits with the man and his wife during the ensuing two years, and a few written notes for another two years.

Family: Both the mother and the father were university graduates. Through their own efforts and achievements they had earned considerable social standing in the community in which they reared their family of three. The patient had two brothers, three years and fourteen years his junior. The family kept together as a quite closely knit unit. Judging from the patient's memories, comments and anecdotes, he was not idealizing when he said: "I think that my relationship with my family, although rocky in spots, has been very happy. I'm devoted to them all. . . ." He and his wife returned home for occasional visits.

Thumbnail self-sketch: "Well, I suppose I'm not an unattractive person for most people. I'm generally optimistic, I think. Probably a little on the lazy side when all is said and done. I probably don't work as hard as I ought to work. I have a pretty good head. It isn't the best head in the world, but it's better than average, and I'm really content with it even though I sometimes wish it were better. I think it's good enough and thank God for what I've got. I expect I'll in my own time do a decent job of living. I don't expect to be much better than average. I expect that I'm sort of average, average good, a little higher than average, in about everything. Average good, in the moral sense, average intelligent, average ambitious, and so on. And that my life will pretty much follow out what—this general route that's kind of set for me, by what I am. I'll do all right. I expect that I can have some pretty rugged things happen to me and it won't jar my general optimism too much. A lot of things I can think of I wouldn't like to have happen, but even so I imagine I'll get along all right no matter what happens because I manage to—for the reason that I am what I am and it has contributed to my average or slightly above average intelligence and so on and so forth, I am able to slough off a lot of stuff better than not. I don't think I'm too dangerously supersensitive or anything of that sort. I imagine I'll belong in the division of perfectly pleasant and not unuseful existence; I hope I'll be able to raise a batch of children that will be as happy as I think I'll be."

Day-to-day routines: At the time of the interviews, the man was well advanced toward obtaining a doctorate in one of the aesthetic disciplines. During the academic year he worked not only at his own studies, but also as a part-time instructor. Whether in the applied, historical or theoretical branches of his field, his accomplishment and achievement had been recognized as outstanding.

There were times, however, when he was not satisfied that he was concentrating on his work sufficiently. "I am ambivalent," he said, "about working harder and taking life easy. I don't expect to be much better than average; I have always been rather modest about my ambitions and expect it is founded on a certain amount of slothfulness. I probably don't work as hard as I ought to work."

The slothfulness or taking life easy, as he called it, appeared in different guises. For example, he was as he said rather modest in his expressed ambitions for a career, contented to aim at a sure target rather than gamble for a great prize. Again, his account of the course of

a typical day gave supportive evidence of a dilatory tendency, one very common in students, namely putting off assignments until the very last minute. The time thus gained was spent with his wife, or at some extracurricular study, or talking with and entertaining friends who might call. Fellow students were frequent callers, as the couple occupied an apartment near the college.

Easy-goingness was also apparent in the infrequency with which the man became agitated, irritated or angry. "I don't know that I've been really angry since I was a little boy," he declared. "Really right down at the guts angry I don't think I've been for a long, long time. I was considerably annoyed last year when one guy wouldn't give me $100 when I wanted it. But anger is kind of exhausting and I tend to avoid it if I possibly can."

It was not that the man was timid or self-effacing, nor a social isolate. While growing up he had always shared some interests with friends of his own age, especially his brother. "But I didn't make friends very easily," he said of grade school days. "And I wasn't a sportsman at all and I frequently got kind of kicked around because I didn't know how to hit a ball with a bat and that sort of thing. . . . But I never felt that life was giving me the blunt end of the stick." He avoided competitive team sports and gymnasium classes because of his genital condition, but joined various other school groups and clubs.

Before leaving high school, he had begun attending a summer colony workshop. There he made many lasting friendships, and there he later met his wife. His first trip abroad had been in childhood with his family, since which there had been others, including one with his wife.

Religious teaching and Sunday school attendance had not been part of the man's childhood experience. Not until college years did he give serious thought to religious, along with philosophical matters. After he met his wife who was devoutly Catholic, he found "Christianity started to make sense, perhaps because she had impressive, implicit faith, and good sense too. I overcame my family-derived horror of Catholicism," but he was far from dogmatic about his personal credo. With his wife he was a regular church attender.

Intelligence test: Wechsler-Bellevue Intelligence Scale results were as follows: verbal IQ 144; nonverbal IQ 133; full IQ 143. A high standard of accuracy was maintained on each subtest. The overall rating was at the level of very superior.

Somatic growth and appearance: After surgical arrestment of pubertal feminization, virilization was induced and maintained by substitution treatment with androgens. The young man had a straightforward understanding of these medical facts, as will become apparent in the following.

Sexuality: "As far back as I can recall, I was always aware of having a genital peculiarity. In our household we were always free and easy about nudity, so I knew I was different from my father and brother." But in the early years of childhood, "it just seemed that men were the superior sex and they had a better thing to pee with."

That the urinary implications of the anomaly were of major import initially was indicated by the first response to the request for earliest memories: "I remember myself squirting the hose. I think I squirted my father in the process. And it was lots of fun." This memory, dating from the age of two, had been reinforced from a photograph taken on the occasion; "and yet, when I look at it, it kind of brings me back, you know, that wonderful feeling of power you have when you're watering something! Well, it kind of brings back something of being a master in your own domain as you squirt this blasted hose around."

Also remembered were a couple of early childhood incidents when the boy and some playmates set up secret urine receptacles behind a garage and in the basement. There were "peeing contests" on a few occasions, including one in which the little boys were amazed to find that a little girl was able to meet a challenge and "pee into a glass."

Sometimes experimentation became more frankly sexual, as when a small group of boys tried to stick assorted items into their orifices; but there were no mutual explorations. While still a young child, the boy learned not to expose himself, regardless of what the others did. His mother, who was less embarrassed by his anomaly than his father, had always talked to him, explaining that he was different from other boys and had special reason not to get into sex play.

"I took more care as I became more aware of the unconscious meannesses of children," he said, "and I got so that I could go to the toilet almost anywhere and manage it so that no one would see." Only once, in high school, did a boy who presumably had participated in infantile sex play make an oblique reference to his friend's genital anomaly. It was in connection with exemption from gymnasium classes. "I said: 'Oh that cleared up a long time ago,' and closed the subject as quickly as I could, but was really quite upset." One or two close contemporary friends were thought to have vague suspicions "that something is wrong." But apart from medical people, wife and family, "nobody else knows as far as I know; and I wouldn't like anybody else to know. I still have a fear that I would be made fun of, and that it would be talked about."

Sex was not a matter for harsh discipline or secrecy in the family. "I knew very early, about six or so, about the process of birth, about the kittens the cat had and where babies come from. It wasn't until I was in the ninth grade that boys told me about the sexual act. I didn't believe it though, and didn't quite dare ask my father. I finally did," but did not get a direct answer, and was eventually convinced from discussions with contemporaries and from comprehensive reading.

"Of course about then it began to dawn on me that I was singularly unequipped; and from then on it became a more acute worry, although it never bothered me very much. But I was aware of it. Other boys had talked about masturbation which I had never thought of up until that time. I had a hell of a time. I experimented for a year before I found out what was necessary, because the kids I knew used the term jerk off or pull off and I didn't know what physically you did. And I didn't realize you were supposed to have an erection to do this. Then I masturbated more or less continuously until I got married. But I never thought it was right; it seems wrong that you should use your head to get out of a trouble which is by the the direct route of logic in nature unnatural."

At the age of eleven "to my great horror I discovered that I had breasts beginning to develop. This seemed quite a calamity. And it was from that time, after the exploratory operation, that I knew I would never have any children, and that was a kind of continual bother. Mother was quite sure that I would never marry;" and was very upset when, with the first serious love affair at nineteen "I said I was damn well going to get married, somehow. Even at sixteen I was thinking about it. Probably because I'd been told I couldn't ever get married anyway. And I had great moral difficulties because I was undecided as to whether I should marry this girl and whether I was going to give some girl a raw deal. Dad was more reticent about it, not knowing quite what to say; and my doctor didn't want to commit himself. But I went ahead anyway.

"In a sense my life has centered around this problem of getting married in that I have always wanted to get married. It's been one of the things that I was going to do, if it was humanly possible. That has been a controlling factor and a challenge all the way along, from some of my earliest memories. Patsy Jane, I can remember when we moved, I kissed her goodbye and took her pictures. I must have been about seven. And then I had a girl friend after that, who continued to write to me." At high school there were the jealousies and rivalries over girl friends; the love notes; "the big fuss about who would dance with whom at dancing school;" the party games of post office; "and, if you were very lucky, the con-

cession of a kiss." In the early college years there was the first serious affair "with Dorothy. And then the girl who is now my wife."

Of the findings at the exploratory operation "I still haven't got the whole works. I still don't know everything about it. But I've gathered that there'd been some kind of female apparatus in there by mistake and that it had been taken out. I also knew that they were looking for testes; and didn't find them. I guess they found a couple of ovaries or something too. That's about all I know about it."

Some weeks later the man did recall in a casual conversation that he had, at the age of eleven, been asked if he wanted to be a girl. The idea had had no appeal. This knowledge, together with the evidence of enlarged breasts, apparently had more significance than that manifested in conscious awareness, witness the following passage of free association: "We ranged in size the same way as we ranged in age. And nobody could ever figure us out when we were on these trips together. They always thought we were some god-forsaken kind of damned pervert bunch. Nobody, obviously I was, I couldn't be the daughter or, or the son or, or Chloe wasn't Homer's daughter and he as', she couldn't be his wife very easily, and I wa', nobody could figure my relationship to; we even considered scratching the name of the institution off the side of the station wagon."

For the most part, however, the ambiguity of sexual status had been well thought out. Only once, in late childhood had there been "some experimenting with my brother, but we were pretty thoroughly frustrated and disgusted and gave that up as a bad job." It was during the college freshman year that the issue came really into focus. "I got to know vaguely of things about drinking; and some of the queer flabs around." One acquaintance "travelled with a weird group of avant garde writers and knew an awful lot of the fairies around." A couple of homosexual guys in the dormitory committed suicide.

"One night a guy came up into my room when I was lamenting about women—I hadn't heard from Dorothy for some time—and looked at me and said: 'Say well, have you ever thought of trying anything else?' And gee, I about vomited on the spot, not knowing whether this was an offer or what. I began to think that maybe other people thought I was funny or something."

Sex was in mind frequently, "and I would think about all kinds of sex, what kinds there were, and then I would wonder if I was safe or not. Whether I would find myself liking it too much or something. Yet, if I ever started making any image of homosexuality, I could never get myself into it It was always other people. Yet I could always think of myself as a possibility for the game. Physically I might have been the best bet in college for some joker. I presumed that some men might find me rather appealing because, well, I'm not a terribly hairy person, and my femininity, in other words, those things about me which are less masculine, less pronouncedly masculine.

"I was much concerned with my breasts at that time. Now I don't care about them, although I am careful how I dress. But at that time people did say things about them. That's something I hadn't thought of, but that might have had quite a lot to do with it. You see I had this tendency to be very conscious, or subconsciously aware of my breasts; and I suppose I always did make some kind of female association there.

"During all this time I was well into this affair with Dorothy; in which I knew consciously and rationally that I had every reason for feeling that I was perfectly okay because I knew perfectly well that I was attracted to women. I had been sneaking out against family opposition, and flying over to spend the weekend with her. And one of my great undergraduate experiences was sleeping all night in the same bed with her. It was strictly a dry run, but it was great sport.

"The guys at school were all big talkers about women and I let it be known that I'd been

a heller when I was in high school—which of course is very far from true—and was now going steady. There were some very vigorous parties, especially at the summer school resort, but I never had any affairs. I would like to have been around raising cain, but in addition to what vague moral feelings I had about it I knew damn well I couldn't. I was very lucky to have any instincts in that direction, I guess. I was taking testosterone; if I wasn't I don't think I would have given a hoot for anybody. I was also in a sense fortunate in falling in with a crowd that was all straight too. I don't think with my particular background I ever would have been very sympathetic to the fairy groups. It was too black and white like that, my family tradition, although there had never been anything very specific about it. I was lucky to be with a gang that were overtly sex happy and spending a lot of time running around finding it."

The resolution of sexual uncertainties was neatly illustrated in a dream, the only one which he could recall. It was dreamed nearly two years earlier, when he and two fellow instructors from the summer school had visited a man whom they thought homosexual. "I dreamed that I was in a church eating supper—stuffed cabbage leaves or something. Rae was with me and we were sitting with strangers at a table in front of the sanctuary. Homer and Chloe were at a table within the sanctuary. The altar was off in the transept. I looked up and in the first pew was this fairy wearing pearl earrings, lipstick and henna in his hair. He was with someone who looked like his mother and was grinning kind of silly. I laughed and went up and told Homer and Chloe to look at him. I came back and was concerned that Rae wasn't eating anything, so we went somewhere else and had supper together. It was a very happy dream. My big problems were resolved. Chloe and I figured it out. I wasn't a fairy because I was with Rae and was able to laugh at this other guy. And I was still able to talk to Chloe and Homer who were my friends, but who wonder about my marrying a Catholic girl and my feelings about Catholicism. And although Rae wasn't quite happy we went somewhere else and were very happy. I remember thinking kind of guiltily that she wasn't eating, because I was enjoying myself tremendously."

"Rae and I had a devil of a time trying to find out whether it was really all right for us to get married. We worried through that. We figured if we could get through that we could get through anything. So I think we can. Gee that was awful. Well, anyhow, we talked to my doctor and he said he thought it would be okay; and we talked to the Church and they said it was okay and so, with many a prayer, we took the giant step and got ourselves engaged and decided it was foolish to wait after you were engaged, and so we got married. At this point we feel, typically, that we're the luckiest people in the world."

Before being married he underwent a plastic repair of the genitalia so that the penis was more advantageously placed for coitus. "And Rae and I make out very well now, of course we're still just married, but it's a rough day when we don't get to bed at least once. And I consider that pretty normal for the first six months of married life. I've never felt that I was particularly abnormal in respect to the strength of libido, but that I seem to fit in the middle somewhere."

Since the age of twelve he had taken testosterone regularly except for one period of several months during which he felt a lack of energy and "probably a little bit of diminution of libido, but not enough to really worry about. I wasn't conscious of being completely apathetic." Ordinarily, there is plenty of erotic sensation localized in the penis. It erects very automatically. "In fact there was one time when I first started taking testosterone when I just needed to move and uff!—there she was, from the friction of my pants."

There is minimal fluid discharge at ejaculation, "but I have the sensation of ejaculation; I'm quite sure it's the same sensation. It may be greater or lesser intensity, I don't know. At any rate it's entirely satisfying. Of course we have had to experiment quite a bit, to find

the best ways for Rae always to have an orgasm," since the erect penis is only about two inches long. "If she doesn't that makes me very unhappy."

A spontaneous gaitey in their marriage was clearly reflected in a passage of free association: "And we had good fun in bed the other day; gee it must have been two hours. We've felt good ever since." At this point he paused, checked himself, awakened from somnolence, laughed and said he had been thinking about examining his wife and of the day when she may get ambitious enough for fellatio in love making. "Rae's awfully nice. She's really awfully nice. I love her a great deal."

The blank-card story of the Thematic Apperception Test evoked a portrayal of family life around the meal table with the children whom they both took for granted they would rear. A family by adoption was, at that time, uppermost in mind, although the possibility of artificial insemination was not excluded. Some years later, the moral issues of artificial insemination having been thoroughly threshed out, the wife became pregnant and gave birth to a child.

PSYCHOLOGICAL APPRAISAL

To all who knew him, it was perfectly obvious that this man had achieved conspicuous, all-round success in coping with life. Only those who knew his medical record knew of the odds against him. To the world at large he gave no sign of having had to surmount the tremendous obstacles imposed by the contradictions of his genital anomaly. He passed simply as an ordinary male college graduate—one of the more stable and well-adjusted.

The challenge to overcome genital defections was pervasive and of long standing: the man said so directly, as well as in unpremeditated sayings and test responses. In early childhood there had been peeing contests; at puberty a total lack of appeal in the idea of changing to live as a girl; in the teens a nagging uncertainty about genital mechanics but a determined resolution to get married; and finally plastic surgery of the genitalia and an erotically successful marriage.

For all its pre-eminence, sexuality did not blot out all else. Academically the man had always made superior achievement. Yet he disparaged himself as being slothful and lazy, and wondered if he might "fall down on the score of lack of penetrating enough intelligence." He spoke of daydream rivalries with a colleague "who is infinitely quicker in mind than I am, which irks me no end."

It seemed possible that the red-hot fire of this man's endeavor had burned in the service of psychosexual problems, and that it might not glow so intensely in the service of work and aesthetic creativeness. Allowing that one must speculate and not be sure in these matters, it also seemed that the kind of logical, systematic literalness requisite for solving psychosexual problems was incompatible with the flashy erraticness of creative insight, penetrating intelligence and a quick mind.

In discussing personal sexual matters, the man exhibited an astonishing degree of empirical detachment and logical reasoning. He had an accurate fund of information about himself which, presumably, had helped him to look at his

own case with detachment. He had not been kept in the dark without answers to questions. He had not been obliged to jump to conclusions, putting two and two together for himself.

Judging from Rorschach and Thematic Apperception Test responses, he was the sort of person whose attention kept pretty well anchored to clear outlines in perceptual experience. He didn't venture too far with confabulation and make-believe. He described and analysed instead. As most people would say, he was very realistic.

Fantasies and personal myths and symbol did not appear to occupy much of a place in the economy of his mental life. It would be brash to theorize about castration, Oedipus or other complexes, on the basis of evidence collected. Suffice it that the man said enough about his childhood relationship with his mother for the Oedipal attachment considered normal in boys to be attributed to him also, his somatic status notwithstanding. It was not an exaggerated or prolonged attachment. Eventually it gave way to a concern with girl friend relationships and marriage, and with the anatomical problem of adequacy in intercourse.

Erotically, there is not a single doubt about the man's masculine orientation though, for those who seriously measure masculinity with a yardstick of pugnacity and aggressiveness, he will not get full marks. He rated himself as being a person slow to anger. His general behavior and demeanor confirmed his estimate.

This slowness to anger was a lack of aggressiveness in the narrow sense of belligerence. It did not include a lack of confidence and initiative making for tentativeness and hesitancy. It was an absence of attack but not of mastery. It was the substitution of strategy for disorganized fighting. At times it was also removal of oneself from provocateurs so that aggression was not aroused.

Though slow to anger, the man was not, then, inhibited and helpless. His control and restraint did not in this or any other context run to excesses; it was temperate and moderate. Thus vigilance was exercised lest the legitimate secret of his anomaly become public, but the secret was openly and frankly discussed under appropriate circumstances. The quality of vigilance and cautious moderation tempered most of his doings in life. His ambitions were modest, he said, and in fact they were, for he had talent and intelligence which would have justified even grandiose aspirations.

All in all, beyond every possible doubt, this person was psychologically a man. He was fortified with a diplomatic arrogance which adjusted to the human demands of the occasion, yet enabled him to choose and select his standards rather than run with the herd. He was meeting life most successfully without any suspicion of psychopathology. There was every reason to believe that he would continue to do so. His life is an eloquent and incisive testimony to the stamina of human personality.

SUMMARY

Seventy-six hermaphroditic patients manifesting somatic ambisexual anomalies were studied psychologically and their gender role and orientation appraised. Gender role and orientation was compared with the sex of assignment and rearing, and with each of five other variables of sex, namely, chromosomal sex, gonadal sex, hormonal sex, internal reproductive organs, and external genitalia. Gender role and orientation was found to be congruous with the sex of assignment and rearing in 72 of the 76 patients, despite contradiction between this pair of variables and one or more of the other five. It was concluded that the sex of assignment and rearing was better than any other variable as a prognosticator of the gender role and orientation established by the patients in this group. The bearing of this finding on instinct theory in psychology and psychiatry was examined. A case report was given to illustrate how gender role and orientation may be fully concordant with the sex of assignment and rearing, despite extreme contradiction of the other five variables of sex.

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FURTHER STUDIES ON THE METABOLIC FATE OF CHLORAL HYDRATE AND TRICHLOROETHANOL[1]

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In a previous communication, it was shown that when single doses of either chloral hydrate or trichloroethanol (TCE) were administered to man, the percentage of chloral hydrate oxidized to trichloroacetic acid (TCA) as calculated from the TCA in the plasma varied from 40 to 87. However, in another group of individuals when calculations were made from urinary excretion after daily constant dosage lower figures were obtained (1). This suggested that the amount of chloral hydrate converted to trichloroacetic acid was not constant from day to day in the same individual, but might be greater in the earlier days of administration. The following observations were made in order to examine this hypothesis and also to determine further the metabolic fate of chloral hydrate and trichloroethanol when administered daily in constant dosage.

METHODS

The subjects of the present study were either healthy volunteers or patients on the medical wards who were in need of sedation[3]. All were given a dose of either chloral hydrate or trichloroethanol each evening. Gelatin capsules containing 0.5 gram each were used[4]. Just before the administration of the drug a sample of blood was drawn and the collection of a twenty-four hour urine sample started. Determinations were made of the trichloroacetic acid content of plasma and urine, and in most instances of the total trichloroethanol (free and glucuronide) content of urine. The analytical methods used were identical with those previously described (1).

RESULTS

When the concentration of TCA in the plasma becomes constant, an equilibrium has been reached in the sense that the amount formed is now equal to the amount being excreted and/or destroyed. After equilibrium is attained, a calculation can be made of the percentage of the daily dose accounted for by renal excretion as trichloroacetic acid and total trichloroethanol during the following 24 hour period.

The data are given in Figure 1, Tables I through V, and protocols of Experi-

[1] This investigation has been aided by a grant from the U. S. Public Health Service.

[2] E. R. Squibb Fellow in Pharmacology and Experimental Therapeutics.

[3] We wish to thank Dr. William F. Fritz for assistance in completing the experiments on the volunteers. In none of the patients studied was there any evidence of liver or kidney disease.

[4] We wish to thank E. R. Squibb & Sons for supplying us with these capsules.

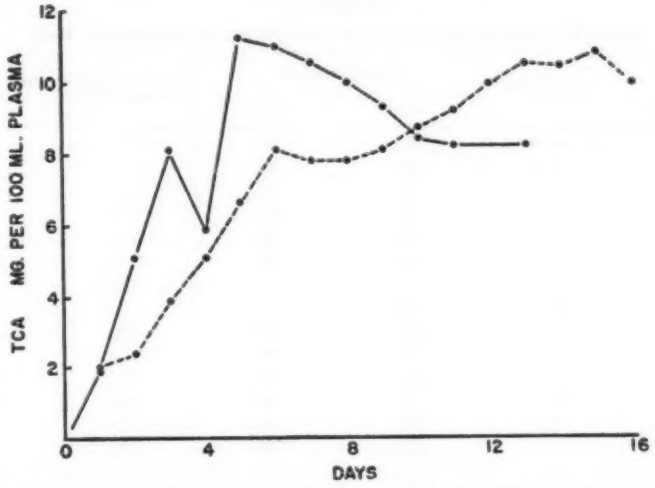

FIG. 1. Plasma concentrations of trichloroacetic acid following constant daily dosage of chloral hydrate. S. C., 140 kg., (○——○) received 1.5 gm. daily and J. K., 73 kg., (○- - -○) 1.0 gm.

TABLE I

Metabolic Fate of Chloral Hydrate in C. B. (71.4 kg.) Receiving 1 gm. Daily

Day	Plasma TCA	Excreted in Urine		Per Cent Dose Excreted as TCA	Per Cent Dose Excreted
		TCA	TCE		
	mg. 100 per ml.	*mg.*	*mg.*		
1	3.5	50	240		
2	5.2	140	295		
3	9.0	234	300		
4	9.9	356	330		
5	10.1	362	396		
6	10.1	337	335		
7	10.4	415	340		
8	10.6	445	368		
9	11.2	460	327		
10	12.0	433	282		
11	12.0	548	260	55	84
12	11.9	670	300	67	100
13	12.0	439	413	44	90
14	12.0	608	348	61	100

ments 1 and 2. Examination of the data shown in Figure 1 and in Table I indicates that the amount of TCA formed from chloral hydrate can vary in the same individual receiving a constant daily dose. In Figure 1, it is to be noted that equilibrium in the case of S. C. with regard to TCA is first reached on days 5, 6 and 7 with about 11 mg. per cent of TCA in plasma, but later on days

TABLE II

Metabolic Fate of Chloral Hydrate in G. B. (68 kg.) Receiving 1.0 gm. Daily

Day	Plasma TCA	Excreted in Urine		Per Cent Dose Excreted as TCA	Per Cent Dose Excreted
		TCA	TCE		
	mg. per 100 ml.	*mg.*	*mg.*		
1	1.4	15	360		
2	3.6	57	455		
3	4.7	76	480		
4	5.3	67	357		
5	6.4	98	365		
6	7.4	—	—		
7	8.3	106	430		
8	8.5	130	430	13	61
9	8.3	130	400	13	57
10	8.3	107	570	11	74
11	8.3	80	430	8	56
12	8.5	122	260	12	41
13	8.3	150	348	15	54

10 to 13, a new equilibrium is attained with only a little over 8 mg. per cent in plasma. Similarly, in J. K., it can be seen that equilibrium is first attained at a concentration of TCA in plasma of about 8 mg. per cent and later is present when the concentration is above 10 mg. per cent. In Table I, the plasma concentrations of TCA indicate a similar situation, equilibrium first being present at 10 mg. per cent and later at 12 mg. per cent.

The amount of TCA excreted in the urine is seen to be variable, not only in different individuals, but in the same individual on different days. Thus, in C. B., Table I, the average excretion of chloral hydrate as TCA is 57 per cent, while in G. B., Table II, it is about 13 per cent. It is difficult to explain the variations in the excretion of TCA in the same individual with its concentration in plasma remaining constant. For example, in Table I, it is seen that the TCA excretion drops from 670 to 439 mg. from day 12 to day 13 with the concentration in plasma unchanged.

An examination of Tables III and IV show that when TCE is administered the amount oxidized to TCA is variable in the two individuals. Thus, the percentages of TCE excreted as TCA are found to average 33 and 4, respectively.

The amount of either chloral hydrate or TCE accounted for by excretion of TCA and total TCE in the urine is very variable in different individuals and in the same individual on different days. Thus, in 2 individuals given chloral hydrate (Tables I and II) the total amount of the daily dose of the drug accounted for in the ensuing 24 hours is on an average 94 and 55 per cent. In another two individuals receiving TCE, the amounts accounted for are 77 and 7 per cent, respectively. The variation from day to day in the same individual is seen in Table II (42 to 74 per cent) and Table III (72 to 92 per cent).

TABLE III

Metabolic Fate of Trichloroethanol in J. B. (76.4 kg.) Receiving 1.0 gm. Daily

Day	Plasma TCA	Excreted in Urine		Per Cent Dose Excreted as TCA	Per Cent Dose Excreteted
		TCA	TCE		
	mg. per 100 ml.	*mg.*	*mg.*		
1	2.1	6	187		
2	6.7	53	316		
3	9.5	75	309		
4	13.5	154	331		
5	14.0	265	312		
6	14.6	218	285		
7	15.9	312	358		
8	16.0	287	370		
9	16.9	398	335		
10	17.1	334	380		
11	18.7	412	485		
12	18.7	340	406	31	72
13	18.7	390	384	35	73
14	18.7	354	600	32	92

TABLE IV

Metabolic Fate of Trichloroethanol in I. H. (80 kg.) Receiving 1.0 gm. Daily

Day	Plasma TCA	Excreted in Urine		Per Cent Dose Excreted as TCA	Per Cent Dose Excreted
		TCA	TCE		
	mg. per 100 ml.	*mg.*	*mg.*		
1	1.3	5	365		
2	3.4	13	370		
3	5.2	34	315		
4	6.1	48	106		
5	6.0	43	32	4	7
6	6.0	38	15	3	5
7	6.0	42		4	
8	6.0	36	53	3	9

Due to the fact that the chlorines of trichloroethanol confer upon the hydroxyl-group something of a phenolic character, we have investigated the possibility of an ethereal sulfate as well as a glucuronide being formed. A sample of the ethereal sulfate of TCE was found to be completely hydrolyzed by boiling for one hour with half normal hydrochloric acid[5]. When a long reflux condenser was used no TCE was lost.

Two individuals were given a constant daily dose of chloral hydrate until equilibrium in the production and elimination of TCA was attained as shown by plasma analyses for TCA. Daily total urine collections were analyzed and the total recovery of chloral hydrate as TCA and TCE determined. In each

[5] We wish to thank Mr. W. A. Lott of E. R. Squibb & Sons for furnishing us with this sample.

TABLE V

Renal Excretion of Chloral Hydrate as Ethereal Sulfate

Date	Plasma TCA	Excreted in Urine		Urinary TCE after Hydrolysis	Per Cent Dose Excreted
		TCA	TCE		
	mg. per 100 ml.	*mg.*	*mg.*		
J. J. (80 kg.)*					
9–29–54	9.1	453	652	640	79
9–30–54	9.5	433	550	542	70
W. J. (71.3 kg.)*					
9–29–54	8.9	175	340	340	37†
9–30–54	9.0	162	425	430	42

* Received 1.5 gm. chloral hydrate daily since 9–21–54.

† About 10–15 per cent loss of urine.

instance recovery was incomplete. Determination of the total TCE before and after hydrolysis gave identical values. Therefore, we can conclude that no ethereal sulfate or other readily hydrolyzable conjugate was present. The data are summarized in Table V.

The finding of a much higher concentration in bile than that in blood of the highly water soluble glucuronide of morphine (2) led us to examine the excretion of trichloroethanol glucuronide in the bile. Our information is based entirely on experiments in dogs.

Two animals were anesthetized with pentobarbital, 25 mg./kg. given intravenously. The common bile duct was then cannulated and the cystic duct ligated. Trichloroethanol, 50 mg./kg., was administered intravenously at zero time and samples of bile and plasma were obtained subsequently. Both renal pedicles were ligated during the experiments in an effort to demonstrate an increased elimination of trichloroethanol glucuronide in the bile. Bile flowed under these conditions at a rate approximating 8 ml. per hour. The following two protocols illustrate these experiments.

Experiment 1. Dog, 11.6 kg. TCE injected intravenously at 0 time. One hour and twenty minutes later both renal pedicles ligated.

Time	Plasma		Bile	
	Free	Total	Free	Total
min.	*TCE, mg. per cent*			
40	1.5	4.4		
60	0.9	4.6	3.0	230
120	0.2	8.3		
145			0.9	210
200	0.0	8.9		
260			0.2	75

Experiment 2. Dog, 13.0 kg. TCE injected intravenously at 0 time. Both renal pedicles ligated at 90 minutes.

Time	Plasma		Bile	
	Free	Total	Free	Total
min.	*TCE, mg. per cent*			
30	1.0	3.6		
80	0.8	6.2	0.2	300
120	0.7	4.6		
150			0.7	224
160	0.2	3.3		

DISCUSSION

This investigation would appear to establish that when chloral hydrate is given in continued daily doses the percentage oxidized to TCA varies not only in different individuals but also in the same individual, that in different individuals receiving daily doses practically all or very little of a given dose of chloral hydrate or TCE can be accounted for by the excretion of TCE and TCA in the urine, that no ethereal sulfate of TCE is excreted, and that trichloroethanol glucuronide is secreted and concentrated in bile. Variation from day to day in the amount of TCA excreted in the urine when the concentration of this substance in the plasma is constant would appear to indicate that the "TCA Space" is variable from day to day, that TCA is excreted by another route than the kidney, and/or that TCA is destroyed in variable amounts from day to day. We have no data to test these possibilities. It is surprising that in some individuals one can account for nearly the entire dose of chloral hydrate or TCE as TCA and total TCE excreted in the urine, while in other individuals only a small fraction of the total dose is accounted for in this way. Obviously, there are for these drugs one or more metabolic routes which are not known and which vary quantitatively from individual to individual. We do not know what percentage of given daily dose of drug is excreted in the bile.

In a previous investigation based on single doses of chloral hydrate and TCE, calculation from the total TCE excreted in the urine, and the TCA and TCE concentrations in plasma appeared to indicate that most of the chloral hydrate and TCE is accounted for by TCE-glucuronide and TCA (1). In the present investigation, in some individuals our findings are in accord with the above experience, but in others one can account for only a small fraction of the dose. This difference in recovery encountered in the two situations may be due to a difference in metabolism of the drug, to inaccuracies in the method of calculation from single doses, or to individual variation.

SUMMARY

When chloral hydrate and trichloroethanol are given in constant daily dosage, the percentage oxidized to trichloroacetic acid can vary in the same individual on different days. In some individuals nearly all of the administered dose may be excreted in the urine as trichloroethanol, trichloroethanol glucuronide and trichloroacetic acid, while in others only a small percentage of the dose is excreted as these substances. The ethereal sulfate of trichloroethanol is not present in urine after administration of chloral hydrate. Trichloroethanol glucuronide is secreted and concentrated in bile.

We wish to thank Sara Ann Verplanck for technical assistance.

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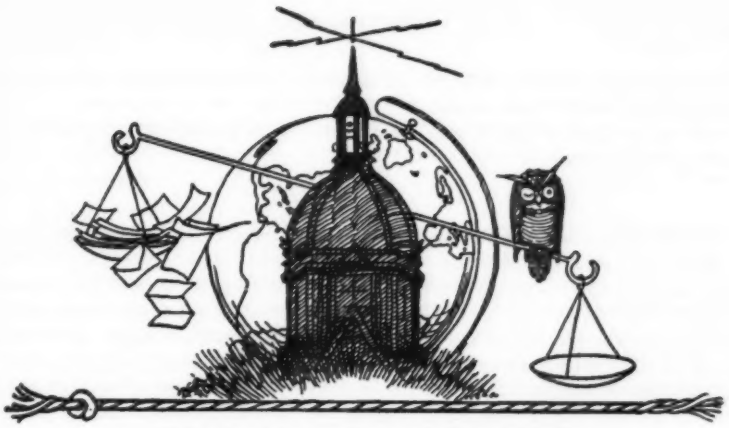

BOOK REVIEWS

Some Physiological Aspects and Consequences of Parasitism. Edited by WILLIAM H. COLE. 90 pp., $2.00. Rutgers University Press, New Brunswick, N. J.

The study of animals and plants which live at the continued expense of other individual hosts is a part of biology. Yet an application of the methods and thinking of modern biology to an analysis of parasitism has been slow in coming. The essays included in this symposium demonstrate that the time is here for such study, and that much can be done. The symposium was apparently organized by Stauber who also contributes a chapter on Leishmaniasis. This facultative intracellular parasite was followed as it multiplied within the cells of the liver and spleen of hamsters. A logarithmic rate of growth occurred shortly after inoculation. The more parasites that were inoculated, the sooner the death of the host, but the time to death was not reduced below thirty days. From this and other data, Stauber emphasizes the need to know more about the effect of the parasite on the host—how the serum protein changes are brought about, etc.

Trager summarizes his studies on the extracellular growth of an intracellular parasite—the plasmodium of bird malaria. He has shown that some of the "reasons" for the "obligate" intracellular growth of this parasite are: a requirement for potassium concentration, and the presence of ATP, along with the unidentified substances in an extract of red cells.

Moulder who has himself investigated the metabolism of both malaria parasites and psittacoid viruses makes a bold and at the same time careful survey of the comparative biochemistry of parasitism. In this he relies heavily on the current model of bacteriophage, but includes also rickettsiae and malaria. The smaller the agent the greater the dependence, and the more complex morphologically the parasite—the greater is its "metabolic competence and independence."

Read analyzes some of the relations of the highly specialized cestodes to their hosts. The processes of excystation are in some cases dependent upon the combined effect of pepsin, trypsin and bile salts. An apparent greater utilization of starch than glucose by the worms living within the intestine seems traceable to a greater period of exposure *in vivo* to the constantly available glucose of the intestinal contents when starch is fed. The peculiar absorptive capacity of the worm through the cuticle is correlated with an extremely complex structure as seen with the electron microscope.

Beuding's studies on the enzymatic and antibody specificity of extracts from schistosomes as compared to muscle give another glimpse of the dependence of agent on host. Taliaferro's

studies of antibody formation although not directly addressed to his old interest in immunity to parasites, are basic to that subject.

These essays mark a beginning of what Stauber calls the log phase of activity in the physiology of parasitism.

FREDERIK B. BANG

The Skin. A Clinicopathologic Treatise. By ARTHUR C. ALLEN. 1048 pp., $25.00. The C. V. Mosby Co., St. Louis, Missouri.

This large profusely illustrated book (1048 pages and approximately 2000 pictures) is a worthy addition to the subject of histopathology of the skin. The author is a general pathologist who, for many years, has had a special interest in dermal pathology. In collaboration with his wife, Dr. Sophie Spitz, he has made distinguished contributions to the problem of pigmented nevi and malignant melanomas by clarifying the concept of the "juvenile melanoma." In Chapter XXIV: Nevi and Malignant Melanomas their views are presented in an attractive, convincing, well documented fashion. During World War II Dr. Allen was assigned to the Armed Forces Institute of Pathology where he took full advantage of the opportunity to draw on one of the largest sources of pathologic material in the world. In 1948 he was requested to select the illustrations for the Institute's *Atlas of Dermatology*. This material provided him with many of the clinical photographs as well as the microscopic sections which appear in this treatise. The reviewer was pleased to recognize several photographs of his former patients at the William Beaumont General Hospital (1944–46). The author also had the good fortune to have had his general work in pathology supplemented by experience as a pathologist in the Tumor Division of the New York Skin and Cancer Clinic in the early forties. His expert knowledge of tumors in all locations is reflected in the high quality of his discussions of skin tumors. In this area he excels. Being a general pathologist, Allen has a comprehensive viewpoint and is able to emphasize related pathology and to include descriptions of general pathology where indicated in such diseases as lupus erythematosus, dermatomyositis, and scleroderma.

MAURICE SULLIVAN

Cold Injury. Transactions of the Third Conference February 22, 23, 24 and 25, 1954. Edited by M. IRENE FERRER. 226 pp., $4.50. The Josiah Macy, Jr. Foundation, New York, N. Y.

The Third Conference on cold injury was held under the auspices of The Josiah Macy Foundation at Fort Churchill, Manitoba, Canada in February of 1954. Two of the participants, M. F. Coffey and J. Le Blanc, were attached to the Canadian Defense Research Northern Laboratories at Fort Churchill. The other participants were transported by air from the United States and Canada. Participating were Drs. John H. Talbott, Albert R. Behnke, George E. Burch, Alan C. Burton, Steven M. Horvath, Harris B. Shumacher and thirteen others interested in the physiology of cold.

A few of the topics presented for discussion were Metabolic Studies of the Eskimo, Diet and Survival, and the Roles of Ascorbic Acid and Subcutaneous Fat in the Prevention of Cold Injury in Man. Experiments employing manual dexterity tests at low temperatures were cited which emphasized the superiority of Indians and Eskimos native to the Arctic over Canadian soldiers. The reasons for this difference remain obscure but it seems likely that in addition to circulatory adjustments there may be more basic biophysical changes in tissue and possibly joint fluid. The problem of acclimatization to cold was discussed and a large mass of animal data presented. The role of ascorbic acid was explored and stimulating observations presented, which suggest that ascorbic acid deficiency renders the human being more susceptible to cold injury.

The goal of The Macy Foundation Conferences is achieved in this volume. Recent developments and investigations not yet completed are discussed by the recognized experts in the field. A certain basic knowledge of cold physiology is assumed and this book is not intended to serve as an introduction and review of the literature in the field of cold injury.

RICHARD S. ROSS

Physiology and Biochemistry of the Skin. By STEPHEN ROTHMAN. 741 pp., $19.50. The University of Chicago Press, Chicago, Illinois.

The reviewer welcomed the publication of this book and used it during the past year as a text for a course on Physiology and Biochemistry of the Skin for graduate students. An opportunity was thus afforded for much closer scrutiny of the contents than is usually the case in the average book review. At the completion of the course my verdict is that Dr. Rothman has produced a classic. This is not only the best recent book on dermatology, it is probably one of the best medical texts of this generation.

The author, a student of Albert Jesionek, Franz Hofmeister, Karl Spiro and Franz Tangl, arrived in America in 1938 and shortly after became associated with the University of Chicago. Taking full advantage of his new opportunity Rothman gathered about him a small band of stalwart young men and inspired them to pursue many of the problems of investigative dermatology which he had conceived in Europe. Several of his original group of fellows and residents collaborated on the preparation of this book and all of them have made excellent contributions. I recall a meeting of the Society of Investigative Dermatology in Atlantic City several years ago when Dr. Rothman and five of his young men motored from Chicago in an old crowded automobile and roomed together in a dormitory of one of the less pretentious hostleries on a side street in order to stay within a budget which was set up to take care of the expenses of one man, namely the Professor. Dr. Rothman gladly exchanged the comforts of a pullman and a luxurious boardwalk hotel for the satisfaction of hearing his five young men present the outstanding papers at the meeting. This is one of many stories that could be told about this kindly man who has taken his rightful place with the leaders of dermatology in the United States. The high quality of his book was no surprise to those of us who know him. It was expected.

In his Foreword Rothman points out that: "Basic research in dermatology is a young branch of the clinical sciences. I have witnessed its rapid development during the last twenty-five years. When, about twenty-five years ago, I first attempted to review all available data on the biochemistry of the skin, my bibliographic studies often were frustrating because of scantiness of pertinent contributions. It was not much better five years later. By now the situation has become reversed. The material has become overwhelmingly large. In preparing this book, particularly during the last two years, I opened with misgivings and uneasiness the newly arriving issues of biomedical periodicals, because their appearance often meant that additions and corrections had to be made in chapters which were by then 'completed.'" Because the material on physiology and biochemistry of the skin is now so immense some selection had to be made. Emphasis was placed on the unique functions of the skin, such as sweat secretion, keratinization, sebum formation and melanin formation and also on the more general functions, in which the skin participates in a particular way, such as cutaneous circulation and vascular reactions. Another principle of selection was to feature those aspects of physiology and biochemistry which seemed to be most important for basic research in dermatology. An effort was made to minimize and/or omit such subjects as the biochemistry of the nucleoproteins which have not been shown to behave differently in the skin from the way they do in other organs.

The chapters on physiology and biochemistry have been integrated rather than presented

separately. The following subjects are presented: Mechanical Properties, Electrical Behavior, Percutaneous Absorption, Circulation and Vascular Reactions, Sensory Functions, Sweat Secretion, Composition of Eccrine Sweat and Aqueous Surface Film, pH of Sweat and Skin Surface, Insensible Water Loss, The Role of Skin in Thermoregulation, The Sweat-Retention Syndrome, Sebaceous-Gland Excretion, Composition of the Lipid Surface Film, Nitrogenous Constituents: General Data, Epidermal Proteins, Collagen, Reticulin, and Elastin, Connective-Tissue Ground Substance, Carbohydrates, Lipid Constituents, Water and Electrolytes, Pigmentation, Enzymes, Carbon Dioxide Delivery, Biology of Epidermal Cells, Hair Growth, Nutritional Influences, and Pathophysiology of Blister Formation.

There is remarkably little repetition in spite of the overlapping nature of the subjects discussed. Because of the concentrated nature of the material some of the chapters required re-reading several times before they were entirely comprehensible to the graduate students and the reviewer who used the text for the course previously mentioned. There are the inevitable few inaccuracies and controversial statements which fail to escape first editions. These are too few and too minor to count.

The bibliography is extensive and can be used to great advantage by all students of medicine. Text books on physiology notoriously neglect consideration of the skin. Now there is a companion text for students of physiology where rapid reference may be made to the multitude of important functions of the skin.

MAURICE SULLIVAN

Diseases of the Skin. For Practitioners and Students, 4th edition. By GEORGE CLINTON ANDREWS. 877 pp., $13.00. W. B. Saunders Co., Philadelphia, Pa.

This is a completely revised and modernized fourth edition of a popular American text. The author, who is the Clinical Professor of Dermatology at Columbia University, is one of the most talented and accomplished physicians in his field. His contributions during the past thirty years have been important and numerous and are illuminated in thirty-eight concise chapters. He is perhaps best known for his investigations on roentgen-ray and radium therapy and the chapters on these subjects are therefore especially noteworthy.

Andrews' new edition is designed for practitioners as well as students. It is attractively presented and should prove to be as popular as previous editions.

MAURICE SULLIVAN

An Atlas of Musculoskeletal Exposures. By H. F. MOSELEY. 235 pp., $22.50. J. B. Lippincott Co., Philadelphia, Pa.

This atlas is a definite help to the surgeon who must know his way into the musculoskeletal system. It deals with the anatomical exposure or approach in the problems which most frequently confront the orthopaedic surgeon.

The author combines the unusual artistic ability of Helen T. MacArthur, whose work is familiar to all, with appropriate photographs and word descriptions of technique.

I expect to use this atlas in my continuing attempts to master the relationship of one anatomical structure to another in the musculoskeletal system.

W. RICHARD FERGUSON

BOOKS RECEIVED FOR REVIEW

Allergy and Anaphylaxis. By KEIZO NAKAMURA, Prof. of Dept. of Bacteriology, Nippon Medical School, Tokyo; Chief of Department of Serology, National Institute of Health, Tokyo, Japan. 114 pp.

A Textbook of Medicine, 9th edition. Edited by RUSSELL L. CECIL and ROBERT F. LOEB. 1786 pp., $15.00. W. B. Saunders Co., Philadelphia, Pa.

Blood Supply and Anatomy of the Upper Abdominal Organs. With a Descriptive Atlas. By NICHOLAS A. MICHELS. 581 pp., $24.00. J. B. Lippincott Co., Philadelphia, Pa.

Collected Papers of The Mayo Clinic and The Mayo Foundation. Edited by RICHARD H. HEWITT, A. B. NEVLING, JOHN R. MINER, JAMES R. ECKMAN, M. KATHERINE SMITH, CARL M. GAMBILL, FLORENCE SCHMIDT and GEORGE G. STILWELL. Vol. XLVI. 843 pp., $12.50. W. B. Saunders Co., Philadelphia, Pa.

Differential Diagnosis. The Interpretation of Clinical Evidence. By A. MCGEHEE HARVEY and JAMES BORDLEY, III. 665 pp., $11.00. W. B. Saunders Co., Philadelphia, Pa.

Experimental Design and Its Statistical Basis. By D. J. FINNEY. 168 pp., $4.50. The University of Chicago Press, Chicago, Ill.

Pain. Its Mechanisms and Neurosurgical Control. By JAMES C. WHITE and WILLIAM H. SWEET. With assistance in the psychiatric sections of Chapters IV and X from STANLEY COBB and FRANCES J. BONNER. 736 pp., $17.50. Charles C. Thomas, Springfield, Ill.

Pathology for the Surgeon, 7th edition. By WILLIAM BOYD. 737 pp., $12.50. W. B. Saunders Co., Philadelphia, Pa.

Perspectives and Horizons in Microbiology. By SELMAN A. WAKSMAN. 220 pp., $3.50. Rutgers University Press, New Brunswick, N. J.

Problems of Consciousness, Transactions of the Fifth Conference, March 22–24, 1954. Edited by HAROLD A. ABRAMSON. 180 pp., $3.50. Josiah Macy, Jr. Foundation, New York, N. Y.

Proceedings of the Forum Sessions, 40th Clinical Congress of the American College of Surgeons, Atlantic City, N. J., November 1954. HARRIS B. SHUMACKER, Jr., Chairman. 851 pp., $10.00. W. B. Saunders Co., Philadelphia, Pa.

Rauber-Kopsch Lehrbuch und Atlas der Anatomie des Menschen, 19th edition. By FR. KOPSCH. Vol. II. 768 pp., Ganzleinen DM 64.50 ($15.35). Georg Thieme Verlag, Stuttgart, Germany. Intercontinental Book Corp., New York, N. Y.

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Vitamin E: Annotated Bibliography 1952 to 1954. Compiled by PHILIP L. HARRIS and WILMA KUJAWSKI. 182 pp., $3.00. The National Vitamin Foundation, Inc., 150 Broadway, New York, N. Y.

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